

Concomitant morphea and lichen sclerosus et atrophicus in the same plaque at the site of intramuscular drug injection: an interesting case presentation

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Abstract

Morphea and lichen sclerosus et atrophicus (LSA) are two diseases that show considerable clinical and histopathological similarity and have been known to coexist in the same patient. Whether the two conditions are different entities or part of the same spectrum has been a topic of debate. This article describes a very rare and interesting case of concomitant morphea and LSA in a 50-year-old female in the same plaque following intramuscular drug injection in the deltoid region of the right arm. The coexistence of morphea and LSA in the same lesion has rarely been reported, thus compelling us to report this case.

Keywords: intramuscular drug injection, lichen sclerosus et atrophicus, morphea, morphea–lichen sclerosus overlap

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Introduction

Morphea is a chronic disease characterized by well-defined plaques with violaceous borders and skin induration. Lichen sclerosus et atrophicus predominantly affects the genital areas and presents with ivory-white shiny papules and plaques with prominent atrophy and telangiectasia. The two conditions may show considerable similarity in clinical and histopathological features to warrant classification as distinct entities (1). However, the two conditions may occur in the same patient at different sites. There have been only a few reports of these two conditions occurring in the same skin lesion, implying that the two conditions may belong to the same disease process. We report a case with features of both morphea and lichen sclerosus occurring in the same plaque following intramuscular injection. The relationship and differences between the two conditions are also discussed.

Case report

A 50-year-old female presented with asymptomatic skin changes on the right arm for 3 months. She had received an intramuscular drug injection in the right arm 3 months prior to the appearance of the skin lesion. The injection that she had received for a fever could not be identified. About 4 days after the injection, the patient developed a raised nodular lesion at the site that gradually flattened and started spreading progressively. Upon examination, the patient was found to have a plaque measuring 13 cm × 5 cm on the right upper arm in the deltoid region, extending from 2 cm below the shoulder joint to the mid upper arm. Morphologically, two types of areas could be distinguished in the plaque. The upper two-thirds were depressed with areas of follicular plugging. The lower third of the plaque was smooth, atrophic, shiny, and wrinkly with areas of hypopigmentation (Fig. 1a). Upon dermoscopy, follicular plugs were seen in the upper part of the lesion but were absent in the lower part (Fig. 1b). The rest of the cutaneous examination, including the genitals and nail fold capillaroscopy, was unremarkable. A systemic examination showed no abnormality.

Laboratory investigations including blood counts and se-

rum biochemistry were within the normal range. Blood glucose levels were within normal limits. Antinuclear antibody, anti-single-stranded DNA antibody, anti-histone antibody, and anti-topoisomerase-11α antibody were negative. Investigation for rheumatoid factor was unremarkable. Thyroid function tests including anti-thyroglobulin antibodies were within normal limits. Serology for *Borrelia* was negative. Skin biopsy was obtained from both the regions of the plaque (the upper two-thirds and lower third). The upper portion showed flattening of the epidermis, basal cell vacuolization, atrophy of the rete ridges and follicular plugs, and a mononuclear inflammatory infiltrate in the papillary dermis consistent with the diagnosis of LSA (Fig. 2). Biopsy of the smooth shiny lower portion showed normal epidermis with thick homogenized collagen bundles and a sparse infiltrate throughout the dermis with adnexal structures trapped deep in the dermis, supporting the diagnosis of morphea (Fig. 3). Based on the clinical and histopathological characteristics, a diagnosis of morphea and LSA in the same plaque was made.



Figure 1a | A single plaque on the right upper arm. The upper two-thirds of the plaque show areas of follicular plugging (black arrows) whereas the lower third is smooth, shiny, wrinkly, and atrophic with areas of hypopigmentation (blue arrows). The site of intramuscular injection is visible.

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Figure 1b | Dermoscopy showing follicular plugging (black arrows) in the upper two-thirds of the plaque.

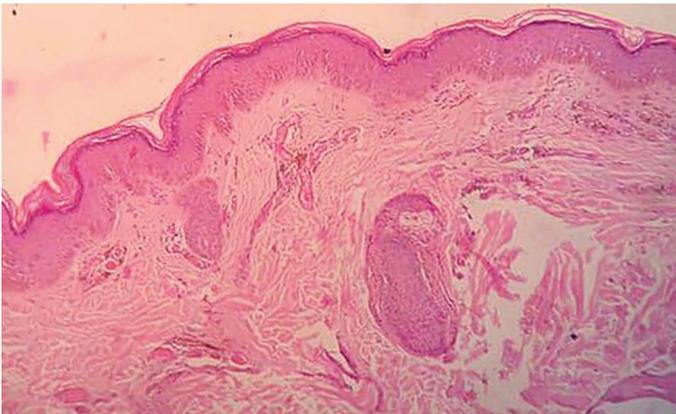


Figure 2 | Histopathology of the upper part of the plaque showing flattened epidermis, follicular plugging, colloid bodies, a hyalinized band of dermis beneath the epidermis, and an inflammatory infiltrate in the superficial dermis (H&E, 10 \times).

Discussion

Morphea is a disease of autoimmune origin that clinically presents as a well-defined shiny, indurated plaque with lilac borders. The histopathology shows inflammation in the reticular dermis with thick bundles of homogenized collagen encroaching the subcutis. LSA, on the other hand, is a disorder of unknown cause, which shows lichenoid papules that coalesce to form a plaque and shows prominent epidermal changes such as atrophy, telangiectasia, and follicular plugging. It is devoid of induration. Histopathology reveals hydropic degeneration of the basal cell layer, keratin plugs, edema, and homogenous upper dermal collagen and inflammation in the mid-dermis (1, 2).

The etiology of morphea is still not clear. However diverse triggering factors have been reported in the literature, which include trauma, varicella, immobilization, Bacillus Calmette–Guérin (BCG), vaccinations for measles-mumps-rubella (MMR), diph-

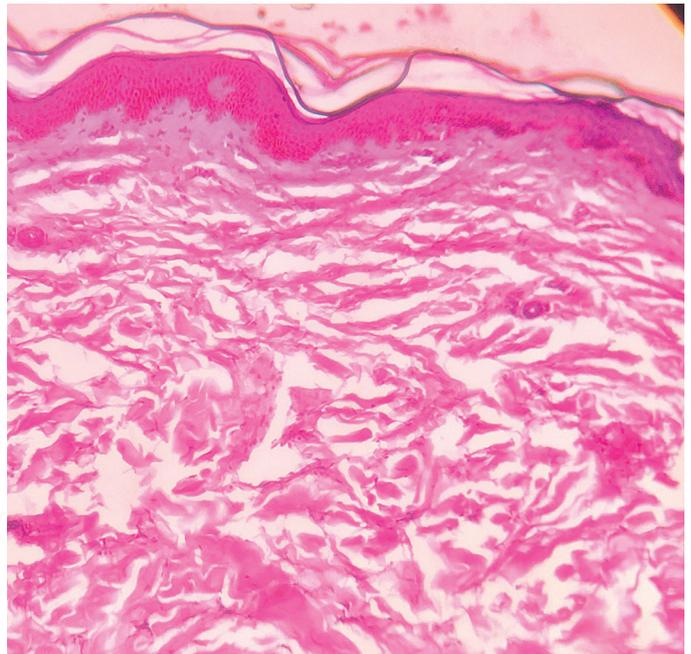


Figure 3 | Histopathology of the lower part of the plaque showing unremarkable epidermis, thick homogenized collagen bundles, and a sparse inflammatory infiltrate with fewer adnexal structures (H&E, 10 \times).

theria-pertussis-tetanus (DPT), and tetanus, injections of vitamin K and vitamin B12, previous radiotherapy, and melphalan limb perfusion. Similarly, LSA has been reported after radiotherapy, trauma, mastectomy, and welding sparks (3–5). However, in our case the combined morphea and LSA occurred at the site of an intramuscular drug injection (the nature of which could not be identified) in the deltoid region.

The two conditions are thus considered separate owing to their distinct clinical and histopathological pictures. However, the differentiation between the two conditions may at times become difficult, both clinically and histopathologically. This is further compounded by reports of the two conditions coexisting in the same patient (6). Some argue that LSA is simply a superficial sub-epidermal form of morphea, and they attribute the edema seen in LSA to lymphatic obstruction due to deep sclerosis in the morphea plaque (7). Uitto et al. postulated that immunological and hormonal factors may be the reason for the varied clinical presentation seen in morphea and LSA despite them being part of a similar disease process. They found that the lesions shared clinical and histological features of both diseases in all 10 of their patients (8).

There have been very few reports of the coexistence of the classical clinically identifiable lesions of morphea and LSA in the same plaque (7, 9). These reports, including the case presented here, support the view that morphea and LSA might be two different clinically diverse diseases, but are probably part of the same disease spectrum. This coexistence occurring at the site of an intramuscular drug injection further adds to the rarity of our case, compelling us to report it.

Conclusion

Although there exist clinical and histopathological differences between morphea and LSA, they arise as a result of the same disease process and should be kept in the same disease spectrum. Keeping both of these disease entities in the same classification system may be justified.

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