Multiple cutaneous reticulohistiocytomas successfully treated with topical psoralen plus ultraviolet A therapy combined with intralesional injections of triamcinolone acetonide

Adam G. Harris, MBChB,[a,b] Asri Wijayanti,[a,b] Marcia Llewellyn, FAAD,[d] Jihan Muhaidat, MBBS,[c] Steven Kossard, FACD,[c] and Dedee F. Murrell, FACD[a,b]
Sydney, Australia

Key words: cutaneous histiocytosis; multiple reticulohistiocytoma; non-Langerhans cell histiocytosis; psoralen plus ultraviolet A therapy; triamcinolone acetonide.

INTRODUCTION

Cutaneous reticulohistiocytosis is a rare form of non-Langerhans cell histiocytosis.[1] Zak[2] originally coined the term *reticulohistiocytosis*, which is now used to describe the histologic pattern of multinucleate giant cells and oncocytic macrophages showing abundant eosinophilic, finely granular cytoplasm with a ground glass appearance.[1] Clinically, reticulohistiocytomas represent asymptomatic, yellow to red-brown or grey-white papules or nodules, which tend to be solitary.[3] Solitary reticulohistiocytomas are regarded as benign and are found to occur more commonly in young males but with no predilection as to location.[3] Reticulohistiocytomas are rarely reported to occur in crops, then termed *multiple reticulohistiocytomas*. These crops show no predilection to age, sex, or location and are thought to be benign with the ability to spontaneously regress.[4-6] In contrast, multicentric reticulohistiocytosis is a well-recognized systemic and aggressive form of reticulohistiocytosis, which typically presents in middle-age women with multiple widespread reticulohistiocytomas and associated severe arthritis. This disease can involve multiple organ systems and is usually progressive, necessitating treatment with systemic agents.[7] Unlike solitary or multiple reticulohistiocytoma, multicentric reticulohistiocytosis can represent a paraneoplastic phenomenon, with up to 25% of cases being associated with an underlying internal malignancy.[8]

Abbreviation used:

PUVA: psoralen combined with ultraviolet A

CASE REPORT

We present a 61-year-old white woman of Hungarian background with a 10-month history of a progressive asymptomatic eruption extending from her left clavicle, up the left side of her neck, and including the pre-auricular region. The eruption consisted of multiple erythematous papules and nodules, some of which coalesced to form large plaques localized only to the left side of the neck overlying the sternocleidomastoid muscle (Fig 1). No regional lymphadenopathy was palpable. Her medical history was significant for obesity, type 2 diabetes mellitus, hypertension, hypercholesterolemia, depression, and irritable bowel syndrome. Her regular medications included insulin, amlodipine, and sertraline.

Results of a biopsy showed prominent interstitial epithelioid histiocytes, some showing a ground glass cytoplasm and a patchy lymphohistiocytic infiltrate with few plasma cells (Fig 2). Stains for acid-fast bacilli and fungi were negative. Immunohistochemical stains were positive for CD68, CD163, CD31, and Factor 13a and negative for CD1a, S100, CD34, adipophilin and erythroblast transformation-specific related gene.

Findings for full blood count, serum electrolytes, and liver function tests were all within normal limits.

From the Department of Dermatology, St George Hospital,[a] Faculty of Medicine, University of New South Wales,[b] Skin & Cancer Foundation,[c] and private practice.[d]

Funding sources: None.
Conflicts of interest: None declared.

Correspondence to: Dedee F. Murrell, FACD, Department of Dermatology, St George Hospital, Sydney, Australia. E-mail: d.murrell@unsw.edu.au.

JAAD Case Reports 2015;1:157-9.
2352-5126

http://dx.doi.org/10.1016/j.jdcr.2015.03.006

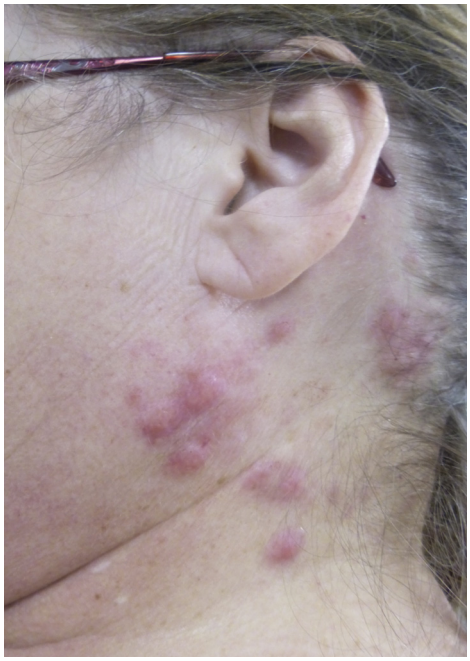

Fig 1. Multiple cutaneous reticulohistiocytomas prior to treatment.

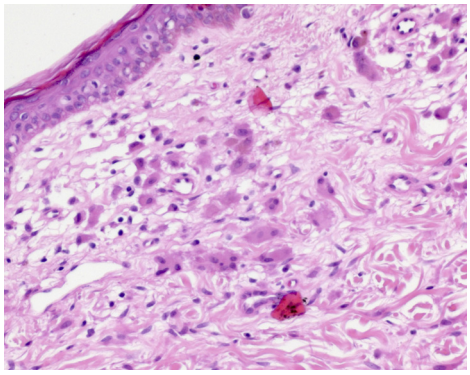

Fig 2. Reticulohistiocytoma, repeat skin biopsy. (Hematoxylin-eosin stain; original magnification: ×20.)

Fasting glucose, cholesterol, and the erythrocyte sedimentation rate were increased, and a small paraprotein of the IgG lambda type was identified in the serum.

A bone marrow trephine and aspirate did not show infiltration of histiocytes into the bone marrow, and computed tomography scans of the chest, abdomen, and pelvis showed no lymphadenopathy but did show some degenerative changes in the left hip. On a clinical and histopathologic basis, a diagnosis of multiple cutaneous reticulohistiocytomas was made.

Treatment consisted of a series of 3 intralesional injections, each containing 10 mg of a 5-mg/mL solution of triamcinolone acetonide given 3 months apart. Immediately after the second injection she was additionally treated with topical application of 0.01% oxsoralen to the lesions followed by psoralen combined with ultraviolet A (PUVA) therapy 3 times a week for a total of 4 months. At the end of the 10-month treatment regimen, there was a marked reduction in the number, size, and height of the papules and nodules (Fig 3).

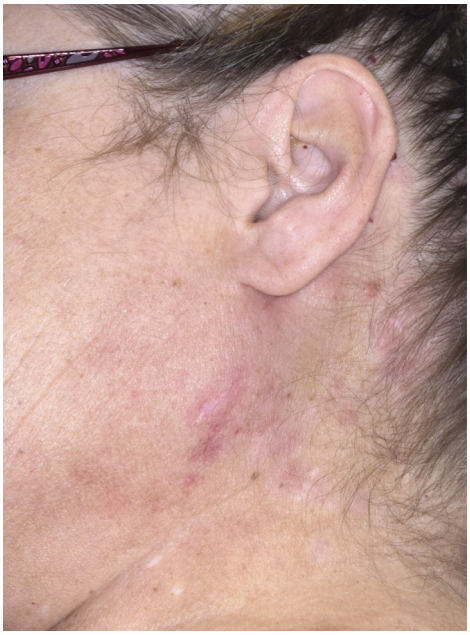

Fig 3. Multiple cutaneous reticulohistiocytomas immediately after treatment with 4 months of topical PUVA combined with intralesional injections of triamcinolone acetonide every 3 months for 9 months.

DISCUSSION

The diagnosis and classification of the different types of non-Langerhans cell histiocytoses are ever changing with the development of new immunohistochemical techniques.[9] There is a paucity of documented cases of multiple cutaneous reticulohistiocytomas of the nonmulticentric type and a very limited number presenting high-quality clinical photographs and potential therapeutic strategies to treat the disease. Multiple cutaneous reticulohistiocytomas have been successfully treated surgically and in combination with electrodesiccation.[5] This treatment is favored for solitary lesions[3] but was difficult to justify in our patient because of the associated morbidity of surgery on the head and neck to remove multiple lesions and because the disease is generally regarded as benign with the possibility of spontaneous regression.[5] Topical PUVA therapy has been used successfully in one case of multiple cutaneous reticulohistiocytomas as a noninvasive method of treatment.[10] We used topical PUVA therapy in combination with intralesional injections of triamcinolone acetonide, which led to a significant

improvement. We feel the temporality of the improvement fits with a therapeutic response to the described treatment, but spontaneous resolution cannot be ruled out.

REFERENCES

1. *Lever's histopathology of the skin,* 10th ed. 2009:675-676.
2. Zak FG. Reticulohistiocytoma ('ganglioneuroma') of the skin. *Br J Dermatol*. 1950;62:351-355.
3. Miettenen M, Fetsch JF. Reticulohistiocytoma (solitary epithelioid histiocytoma): a clinicopathologic and immunohistochemical study of 44 cases. *Am J Surg Pathol*. 2006;30:521-528.
4. Zelger B, Cerio R, Soyer HP, Misch K, Orchard G, Wilson-Jones E. Reticulohistiocytoma and multicentric reticulohistiocytosis. *Am J Surg Pathol*. 1994;16:577-584.
5. Toporcer MB, Kantor GR, Benedetto AV. Multiple cutaneous reticulohistiocytomas (reticulohistiocytic granulomas). *J Am Acad Dermatol*. 1991;25(5 Pt 2):948-951.
6. Goette DK, Odom RB, Fitzwater Jr JE. Diffuse cutaneous reticulohistiocytosis. *Arch Dermatol*. 1982;118(3):173-176.
7. Islam AD, Naguwa SM, Cheema GS, Hunter JC, Gershwin ME. Multicentric Reticulohistiocytosis: a Rare Yet Challenging Disease. *Clin Rev Allergy Immunol*. 2013;45:281-289.
8. Snow JL, Muller SA. Malignancy associated multicentric reticulohistiocytosis: a clinical and immunophenotypic study. *Br J Dermatol*. 1995;133(1):71-76.
9. Zelger B, Sidoroff A, Orchard G, Cerio R. Non-Langerhans cell histiocytoses. A new unifying concept. *Am J Dermatopathol*. 1996;18:490-504.
10. Dawe RS, Cameron H, Lowe JG. PUVA for diffuse cutaneous reticulohistiocytosis. *Br J Dermatol*. 1998;138(4):720-721.