Successful treatment of multinucleate cell angiohistiocytoma in an adult male patient with potassium-titanyl-phosphate laser in combination with intralesional corticosteroids

Bryn Edgerton, BSc (Hons),[a] Malika A. Ladha, MD,[b] Charlene Hunter, MD,[c] Alim R. Devani, MD,[d] and Vimal H. Prajapati, MD[b,d,e,f]
Calgary, Canada

Key words: dermatopathology; fibrohistiocytic; general dermatology; laser; multinucleate cell angiohistiocytoma; potassium-titanyl-phosphate; vascular.

INTRODUCTION

Multinucleate cell angiohistiocytoma (MCAH) is a rare, benign, idiopathic disease characterized by strong female predominance and predilection for the extremities and face.[1] Most often asymptomatic, MCAH follows an indolent, yet progressive course, with spontaneous remission being uncommon. Many aspects of this vascular and fibrohistiocytic proliferation are controversial, including pathogenesis and treatment recommendations. We present a case of MCAH in a male patient successfully treated with potassium-titanyl-phosphate (KTP) laser.

CASE REPORT

A 67-year-old man with Fitzpatrick skin type IV was assessed in an outpatient dermatology clinic for 1-year history of an asymptomatic cutaneous eruption. Medical history was significant for gout, hypothyroidism, hypertension, and dyslipidemia. Medications included allopurinol, levothyroxine, losartan, and rosuvastatin. Physical examination found multiple, discrete, well-circumscribed, smooth, somewhat flat-topped, erythematous-to-violaceous papules on the bilateral dorsal hands (Fig 1). Skin biopsy revealed an increased number of vascular channels, fibrohistiocytic cells, and large angulated multinucleate cells within a collagenous stroma in the papillary and mid dermis (Figs 2 and 3).

Abbreviations used:

ILC:	intralesional corticosteroid
KTP:	potassium-titanyl-phosphate
MCAH:	multinucleate cell angiohistiocytoma

A diagnosis of MCAH was favored given the clinical and histopathologic findings.

The patient requested treatment for cosmetic purposes. Initial monotherapy with 2 sessions of intralesional corticosteroid (ILC) injections (0.5 mL of 10 mg/mL triamcinolone acetonide), each 8 weeks apart, provided no improvement. At 16 weeks, he began combination therapy with ILC and KTP laser. The patient received 2 sessions of ILC injections (0.3 mL of 5.0 mg/mL and 0.1 mL of 5.0 mg/mL triamcinolone acetonide) combined with KTP laser (wavelength, 532 nm; fluence, 12.0 J/cm^2; spot size, 5.0 mm; pulse duration, 15 ms), each 10 weeks apart. This treatment was well tolerated without complications and resulted in significant improvement in both induration and discoloration (Fig 4).

DISCUSSION

MCAH is a rare diagnosis; fewer than 150 cases are described in the literature.[1] It is likely underreported because of a lack of physician knowledge regarding this condition. A review of 142 published case

From the Cumming School of Medicine[a]; Division of Dermatology, Department of Medicine[b]; Department of Pathology and Laboratory Medicine[c]; Division of Community Pediatrics, Department of Pediatrics[e]; and the Division of Pediatric Rheumatology, Department of Pediatrics, University of Calgary[f]; and the Dermatology Research Institute.[d]

Funding sources: None.
Conflicts of interest: None disclosed.
This report was presented as an E-Poster at the World Congress of Dermatology 2019.

Correspondence to: Vimal H. Prajapati, MD, Dermatology Research Institute, Suite 330, 8500 Blackfoot Trail S.E., Calgary, Alberta, Canada, T2J 7E1. E-mail: vimal@dermatologyresearch.ca.

JAAD Case Reports 2019;5:880-2.
2352-5126

https://doi.org/10.1016/j.jdcr.2019.08.001

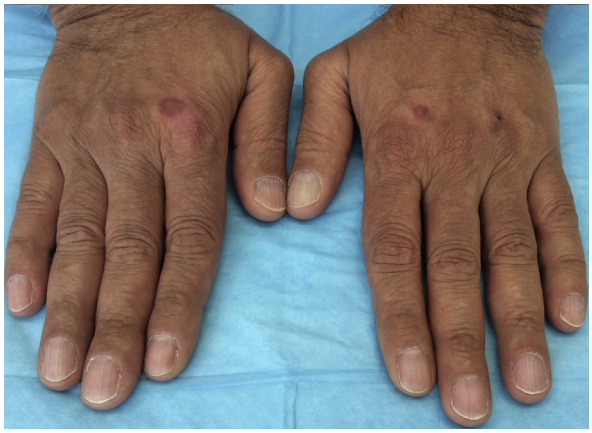

Fig 1. Clinical findings at baseline. Multiple, discrete, well-circumscribed, smooth, somewhat flat-topped, erythematous-to-violaceous papules on the bilateral dorsal hands.

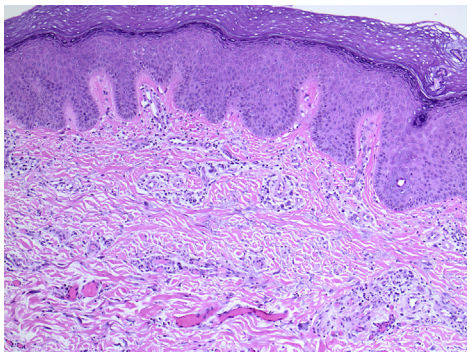

Fig 2. Histopathologic findings at 10× magnification. Numerous small ectatic blood vessels and increased dermal cellularity within a collagenous stroma in the superficial and mid dermis.

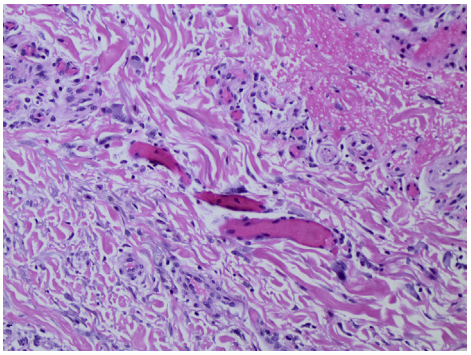

Fig 3. Histopathologic findings at 20× magnification. Numerous scattered large angulated multinucleate cells and fibrohistiocytic cells.

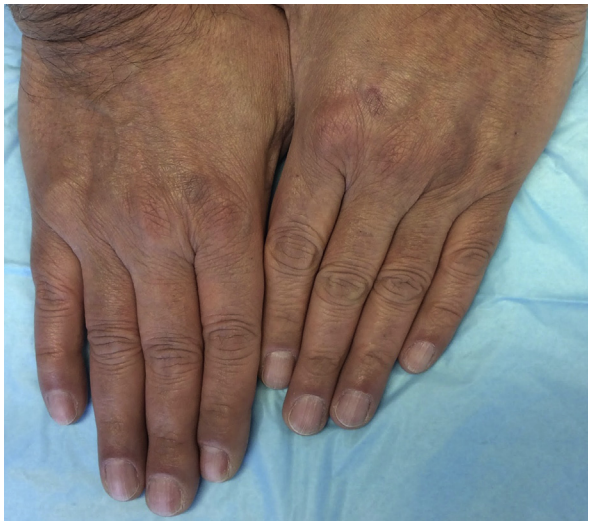

Fig 4. Clinical findings at 8-month follow-up. Significant reduction in the induration and discoloration of MCAH after KTP laser.

reports found that the average age at diagnosis is 50.1 years and women are more commonly affected (79%).[1] Our 67-year-old male patient did not align with either of these trends.

The pathogenesis of MCAH is complex with both a vascular and fibrotic etiology. It is hypothesized that this condition results from an active inflammatory response to intravascular macrophage migration and increased dermal vascularity.[1] In addition, affected areas exhibit an overexpression of estrogen receptor α.[2] Estrogen signaling has been linked to angiogenic effects.[3] This finding potentially explains the highly vascular nature and female predominance of MCAH.[1]

MCAH clinically presents as well-defined, grouped, erythematous-to-violaceous papules, with a predilection for the extremities and face.[1] The clinical differential diagnosis includes granuloma annulare (papular variant), sarcoidosis (papular variant), angiofibroma, dermatofibroma, and Kaposi sarcoma. Given that these are often clinically indistinguishable, a skin biopsy is required. On histopathology, MCAH shows dilated capillaries and small vessels in the dermis, mid-dermal fibrosis with thickened collagen bundles, and the presence of multinucleated cells.[1] The histopathologic differential diagnosis mainly includes angiofibromas and dermatofibromas; increased vascularity and the presence of multinucleated cells distinguish MCAH from the aforementioned entities.

Although most cases are asymptomatic, pruritus may be present.[4] This condition is considered benign; however, spontaneous remission is reported rarely.[5] Patients often seek treatment for cosmetic purposes. Multiple treatments have been reported for MCAH with varying success, including surgical excision, cryotherapy, laser (argon, carbon dioxide, pulsed dye), and intense pulsed light.[4,6-9]

Similar to another case report, our patient did not respond to monotherapy with ILC injections.[6] The sole use of corticosteroid likely did not impact the vascularity; thus, our patient had no clinical improvement with ILC injections. KTP laser was therefore selected to target the vascular component. The latter is found to markedly reduce blood vessels in superficial vascular lesions including, but not limited to, telangiectasia, angioma, hemangioma, and venous lake.[10] The use of KTP laser enabled a reduction of the vascular proliferation of MCAH, resulting in a significant improvement without any adverse effects. This approach can thus be added to the therapeutic armamentarium for MCAH, which is often treatment refractory.

REFERENCES

1. Frew JW. Multinucleate cell angiohistiocytoma: clinicopathological correlation of 142 cases with insights into etiology and pathogenesis. *Am J Dermatopathol*. 2015;37(3):222-228.
2. Cesinaro AM, Roncati L, Maiorana A. Estrogen receptor alpha overexpression in multinucleate cell angiohistiocytoma: new insights into the pathogenesis of a reactive process. *Am J Dermatopathol*. 2010;32(7):655-659.
3. Losordo DW, Isner JM. Estrogen and angiogenesis: a review. *Arterioscler Thromb Vasc Biol*. 2001;21(1):6-12.
4. Perez LP, Zulaica A, Rodriguez L, et al. Multinucleate cell angiohistiocytoma. Report of five cases. *J Cutan Pathol*. 2006;33(5):349-352.
5. Shapiro PE, Nova MP, Rosmarin LA, Halperin AJ. Multinucleate cell angiohistiocytoma: a distinct entity diagnosable by clinical and histologic features. *J Am Acad Dermatol*. 1994;30(3):417-422.
6. Applebaum DS, Shuja F, Hicks L, Cockerell C, Hsu S. Multinucleate cell angiohistiocytoma: a case report and review of the literature. *Dermatol Online J*. 2014;20(5):22610.
7. Kopera D, Smolle J, Kerl H. Multinucleate cell angiohistiocytoma: treatment with argon laser. *Br J Dermatol*. 1995;133(2):308-310.
8. Richer V, Lui H. Facial multinucleate cell angiohistiocytoma: long-term remission with 585 nm pulsed dye laser. *Clin Exp Dermatol*. 2016;41(3):312-313.
9. Fernandez-Jorge B, del Pozo J, Garcia-Silva J, Barja JM, Yebra-Pimentel MT, Fonseca E. Multinucleate cell angiohistiocytoma: treatment using intense pulsed light. *Dermatol Surg*. 2009;35(7):1141-1143.
10. Becher GL, Cameron H, Moseley H. Treatment of superficial vascular lesions with the KTP 532-nm laser: experience with 647 patients. *Lasers Med Sci*. 2014;29(1):267-271.