Case report

Livedoid vasculopathy successfully treated with hyperbaric oxygen

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Abstract

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Livedoid vasculopathy is a painful, ulcerating condition of the lower legs, ankles and feet with the typical histological feature of hyalinising vascular change of dermal blood vessels with minimal inflammation. Therapeutic interventions have been diverse and varyingly successful. We report a biopsy-proven case in a 27-year-old male, which responded rapidly and completely to hyperbaric oxygen therapy. A few such cases have been reported previously, but only in dermatological journals, not in the hyperbaric medicine literature.

Key words

Wounds, hyperbaric oxygen, hyperbaric oxygen therapy, case reports

Introduction

Livedoid vasculopathy (LV) is a painful, ulcerating condition of the legs, especially the ankles and feet. It has had many names, including livedoid vasculitis, segmental hyalinising vasculitis, *atrophie blanche* and PURPLE (painful purpuric ulcers with reticular pattern of the lower extremities).[1-3] It is most common between 15 and 50 years of age and affects females two to three times more frequently than males.[2]

The pathophysiological mechanism is considered to be a vaso-occlusive phenomenon due to intraluminal thrombosis of dermal venules.[2] Typical histological features are of hyalinising vascular changes of the subintimal layer of dermal blood vessels with minimal inflammation.[4] Inherited defects of coagulation, thrombophilias and auto-immune connective tissue diseases have been associated with LV.[2] The typical clinical presentation is the development of painful, purpuric, erythematous, papular plaques and papules which may then become vesicles and ulcerate. These heal slowly over weeks or months leading to white atrophic scars (*atrophie blanche*).[2]

Pain is a constant characteristic and may be debilitating. This pain is perhaps ischaemic in nature as reduced transcutaneous oximetry measurements (TCOMs) have been documented in 20 of 27 patients (74.1%) in one case series.[4] Many therapies have been promulgated, including various regimens of anticoagulation (aspirin, heparin, low molecular weight heparins and warfarin), low-dose tissue plasminogen activator (t-PA), and hyperbaric oxygen therapy (HBOT) has been used in a few patients.[2,5–8] As the literature is very limited, a further case given HBOT is reported here. The patient provided written consent for this report.

Case report

A 27-year-old, previously fit, male, non-smoking geophysicist contacted our hyperbaric medicine unit with a view to HBOT for his ongoing painful foot ulcers from biopsy-proven livedoid vasculitis. The patient had found a report on the internet of the successful treatment of two patients with LV in a dermatology journal and requested a trial of HBOT prior to considering methotrexate, which his dermatologist had advised.[5]

The patient reported developing a "*bruising*" effect on his feet a year earlier, which had spread over a few weeks and become quite painful with some open wounds. This was initially diagnosed as eczema by his general practitioner but as it was unresponsive to topical steroids, he was referred to a dermatologist who diagnosed LV, confirmed on punch biopsies of both feet. He was commenced on prednisolone 25 mg daily for a week and then tapered over two weeks, with a good but incomplete response, with most of the ulcers healing. However, upon steroid cessation the condition returned. Many subsequent courses of steroids over the next year had the same transient effect, with the condition becoming progressively worse such that most of both feet and ankles were covered in ulcers.

Apart from aspirin, 50 mg daily, there had been no trial of other anti-thrombolytic therapy or anticoagulants. Immunological testing for an auto-immune cause was negative as were a thrombophilia screen and serum protein electrophoresis. A full blood count, CRP and ESR were all normal. Bilateral leg arterial Doppler studies were normal with an ankle brachial index (ABI) of 1.1. There was no clinical evidence of varicose veins.

He was assessed as to his fitness for HBOT and no contraindications were present (he had been an enthusiastic scuba diver). TCOMs were not performed. He commenced HBOT (90 min at 243 kPa in a monoplace chamber) six weeks after presentation. Within the first week of his planned six-week course of HBOT (30 sessions) he reported

a dramatic lessening in pain and a reduction in the extent of ulceration. By the end of the course, all of his ulcers had healed for the first time since their original onset.

Upon review six months later, he reported: "*I have absolutely no pain, no ulceration, my scarring has reduced and I am living my life entirely as I was prior to becoming ill*". At recent further contact, over a year after completing HBOT, the patient remained well and without recurrence.

Discussion

There are limited reports of HBOT for treating LV, all of which have been published in dermatology journals. Some of these report success, others failure.[2,5–8] A recent review of LV reported four patients treated with HBOT for 20–25 sessions with complete healing of the lesions. Pain was completely relieved between the seventh and twelfth sessions.[2]

This early relief of pain with subsequent complete healing without relapse, as occurred in our case, was also reported in a series of twelve patients.[5,6] Eight patients completed the planned course of HBOT with reduction in analgesic usage after an average of five HBOT and complete healing at a mean of 3.4 weeks (range 2–5 weeks).[5,6] Six of these patients subsequently relapsed but responded to further HBOT. A transient increase in pain (not evident in our case) was noted in some after their first HBOT, which soon resolved in those that chose to continue. Two patients withdrew from further HBOT after the first session because of this increased pain. The outcome of the four patients that did not commence or complete HBOT in this series is not described. The authors commented that another mechanism of response to HBOT in LV, in addition to that usually attributed to HBOT in wound healing, could be the effect of HBOT on increasing the release of various fibrinolytic (including t-PA) molecules from endothelial cells. This effect of HBO_2 had been reported previously *in vitro* and *in vivo*.[9,10]

A therapeutic 'ladder' for treatment of LV with references (which included HBOT) was published in 2006.[1] HBOT was tenth on the list of 12 considered therapies. There are no randomised controlled trials of any therapy for LV, and all forms of treatment proposed for LV are based on reports of isolated cases or case series.[2]

Conclusion

HBOT should be considered as a therapy in patients with livedoid vasculopathy, particularly where other treatments have failed and where pain is a significant factor and in whom there are no significant contraindications to its use.

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Editor's note:

Colour photos of the patient's feet and ankles just prior to and at the end of the HBOT course may be viewed until 01 June 2013 on the journal website <http://dhmjournal.com>; select "*LV case photos*".