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Case Report

Chronic Regional Pain Syndrome After Subtalar Arthrodesis Is Not Prevented by Early Hyperbaric Oxygen

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Subtalar arthrodesis was performed on a 48-year-old, non-insulin-dependent diabetic with a history of chronic ankle instability and lateral ankle pain. In the early post-operative period he presented as an emergency with an infection at the operative site. This was treated with 2 returns to the operating theatre for washout and debridement. His wounds were left open and at 3 weeks after emergency admission he was referred for adjunctive hyperbaric oxygen (HBO) therapy to aid healing by secondary intention. He received a total of 19 hyperbaric sessions, at a pressure of 2.2 ATA, one treatment per day for 5 days a week.

Shortly after commencing HBO therapy his ankle became increasingly painful, despite the introduction of analgesia. By 7 weeks after emergency admission his wounds had virtually healed but hyperesthesia persisted over the dorsum of the foot. A computerized tomography scan at 5½ months post-operatively showed satisfactory joint fusion and revealed no evidence of infection. Symptoms and signs at this time were compatible with a diagnosis of chronic regional pain syndrome (CRPS).

There is published evidence to suggest that HBO therapy may be a useful modality in the treatment of established CRPS. Here, we seek to publicize a case in which early treatment with HBO for another indication did not prevent the simultaneous development of CRPS Type 1.

Key words: Subtalar arthrodesis, hyperbaric oxygenation, chronic regional pain syndrome

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Following multiple inversion injuries to the left ankle and a soft-tissue (Brostrum) repair for moderate instability 2 years previously, a 48-year-old, non-insulin dependent diabetic presented with significant anterior ankle pain. Intraarticular injection of steroid brought no improvement. Subtalar arthrodesis of the ankle was performed, with clearance of cartilage from the talo-calcaneal, calcaneo-cuboid, and talo-navicular joints and placement of screws in the first 2 of these joints. After an in-patient stay of 4 days he was discharged in a cast for later outpatient review.

He presented unexpectedly to the emergency room one week later with a swollen, painful foot. He was apyrexial, with vital signs within normal limits. His C-reactive protein (CRP) was raised at 60 mg/l and he was taken to the operating theatre for debridement and washout. The metalwork was left in-situ. Culture of intraoperative samples grew staphylococcus aureus. He was started on intravenous vancomycin. Two days later, although he remained systemically well, his CRP was now 133mg/l. A second washout was performed, with evacuation of hematoma. Once again the metalwork was left in-situ. Two wounds on the

dorsum of his foot (each 1 x 2 cm) and a third lateral wound (10 x 3 cm) were left open and a vacuum dressing was applied. After a further 2 days his antibiotics were changed to oral clindamycin, later amended to combination therapy with levofloxacin and rifampicin. By 1 week after his emergency admission his CRP had fallen to 32 mg/l. By 3 weeks after admission it was below 10 mg/l. At this time he was referred for hyperbaric oxygen (HBO) therapy to aid wound healing by secondary intention. Shortly after, as there was no clinical evidence of infection, antibiotic therapy was halted.

Method

Adjunctive hyperbaric oxygen (HBO) therapy was commenced at 3½ weeks after his emergency admission. Measurement of tissue response to hyperbaric oxygenation was not undertaken, there being no prior history suggestive of vascular compromise. The hyperbaric protocol employed is shown in Fig. 1.

Hyperbaric oxygen (100% oxygen at 2.2 ATA) was delivered in a multiplace chamber on the basis of one treatment per day, 5 days per week for a total of 19 sessions. The appearance of his left ankle and foot at the commencement of hyperbaric treatment is shown in Figure 2. A notation in his patient record, made by a senior nurse in attendance at this time, recorded that his pain level appeared high.

Within a week of commencing hyperbaric therapy, his wound was assessed by the plastic surgery team which felt that it was healthy and healing satisfactorily. He was also reviewed at this time by a consultant pain physician who noted a neuropathic component to his ongoing pain and commenced gabapentin and oxycodon. CRP, taken at the midpoint of his hyperbaric treatment, remained below 10mg/l.

Results

Seven weeks after his emergency admission, and despite almost complete healing of the wounds, hyperesthesia persisted over the dorsum of the foot. A month later he attended the pain clinic once more and was again reviewed by the senior consultant pain physician. At this stage his foot was noted to be edematous and violaceous in appearance, with decreased sudomotor activity and muscular wasting. Despite the earlier medications, his pain had worsened and the presence of allodynia was documented. At no point was physiotherapy attempted as the patient's pain precluded any participation. Figure 3 shows the appearance of the foot at this time.

Computerized tomography scanning at 5½ months post-operatively showed satisfactory fusion of the subtalar and calcaneo-cuboid joints and confirmed the presence of osteoarthritic degenerative changes in the talo-navicular joint. No lucent areas suggestive of infection were seen around the metalwork. The patient nevertheless continued to experience constant pain at

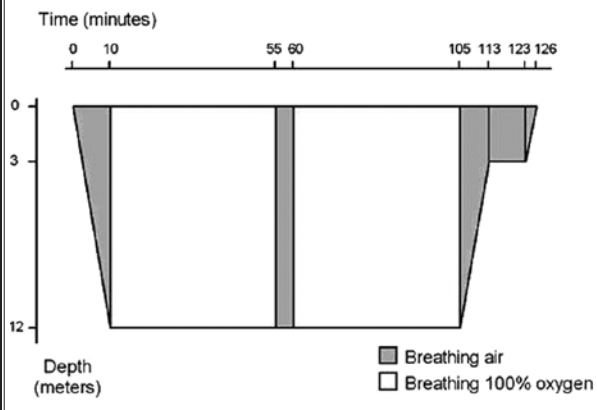

Fig. 1. *The hyperbaric protocol employed.*

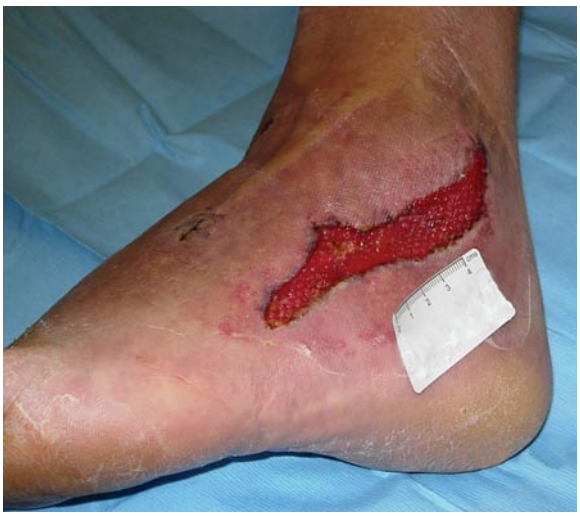

Fig. 2. *Appearance of the left foot and ankle at the commencement of hyperbaric treatment (5 weeks after operation).*

rest, only partially controlled by oral medication, such that he was unable to bear weight on the affected side. A therapeutic sympathetic blockade was offered, but declined. By 11 months the changes in the skin had settled, as shown in Figure 4, though the hyperesthesia remained and the patient was still not able to bear weight. Alternative techniques of analgesia such as spinal cord stimulation were being explored.

Discussion

Hyperbaric oxygenation therapy is a treatment in which the patient breathes 100% oxygen at pressures greater than atmospheric (1). Breathing high concentrations of oxygen at surface pressure and the topical exposure of limbs to high concentrations of oxygen are excluded by this definition. In well-perfused, normally oxygenated tissue, HBO treatment confers no benefit to wound healing (2). In situations where healing is expected to be, or has become problematic, the effects of treatment with HBO at cellular and tissue level may confer useful benefit in terms of increased oxygenation, edema reduction, and potentially enhanced tissue repair. There is sufficient evidence for its efficacy in this approved indication to justify reimbursement by insurers of the costs of treatment (3).

Oxygen diffusion distance through tissue fluids is proportional to the square root of the oxygen concentration in the source capillary (4). At 2 ATA pressure, the diffusion distance of oxygen through tissue fluids is increased by a factor of 3 (2). Oxygen at partial pressures greater than 500 mmHg causes vasoconstriction (5) through contraction of smooth muscle vasculature. Although this is estimated to cause a 20% reduction in limb blood flow (6), any decrease in oxygen delivery attributed to vasoconstriction is more than offset by the increased oxygen tension (2). Upstream vasoconstriction prompts a fall in downstream capillary hydrostatic pressure, reducing capillary transudation and favoring reabsorption from the interstitium. The net effect is a reduction of tissue edema (7).

Processes involved in wound repair are oxygen-dependent. In hypoxic conditions angiogenesis is slowed (8). Hyperoxia enhances angiogenesis (9) and stimulating fibroblast proliferation and collagen synthesis (10). Lack of oxygen also renders wounds liable to infection; when partial pressures fall below 30 mmHg, leukocyte function is compromised (11). HBO therapy enhances the oxygen-dependent, intracellular-killing capability of leukocytes (12) by providing substrate for the formation of oxygen free radicals and augmenting the oxidative respiratory burst (13). The combination of these effects leads to increased rates of wound closure in hypoxic tissues (14).

Complex regional pain syndrome (CRPS) encompasses chronic pain and a range of accompanying symptoms which occurs after injury. The duration and magnitude of the symptoms, however, exceed the clinical course which might be anticipated from the initial event. In CRPS type I, previously known as reflex sympathetic dystrophy and Sudeck's atrophy, the symptoms are initiated by soft tissue injury or fracture

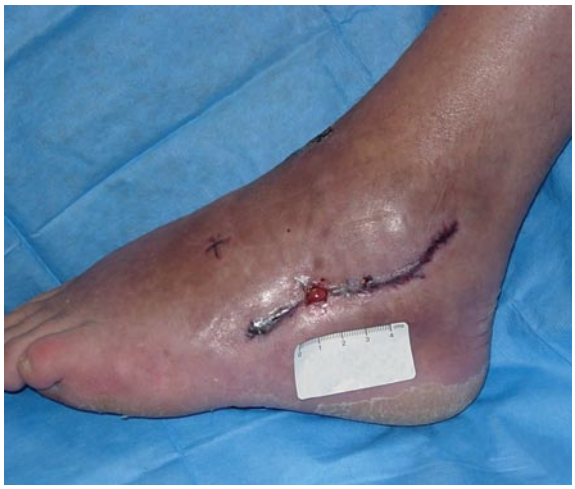

Fig. 3. *Appearance of left ankle and foot at 13 weeks after operation.*

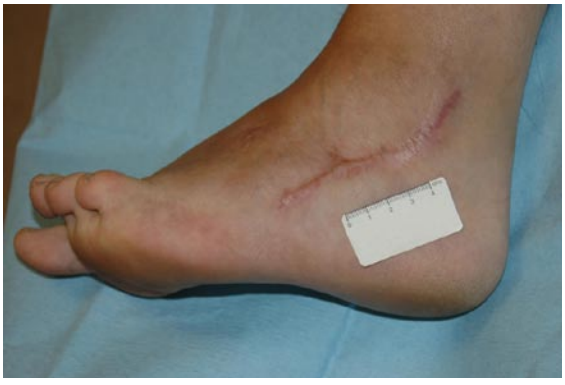

Fig. 4. *Appearance of left ankle and foot at 11 months after operation.*

of a limb. CRPS type II follows injury to a major peripheral nerve (15).

Revised diagnostic criteria for CRPS have been proposed by Norman Harden and Bruehl (16). For a positive diagnosis, subjects should exhibit continuing pain disproportionate to any inciting event, with no other diagnosis that would better explain the sign and symptoms which appear in Table 1. With regard to symptoms, subjects should report at least one symptom in 3 categories for a clinical diagnosis. A research diagnosis requires the report of symptoms in all 4 categories. With regard to signs, both clinical and research diagnoses share the stipulation that one sign must be observed in 2 or more categories at the time of diagnosis. The subject in this case displayed sufficient symptoms and signs for a diagnosis of CRPS Type 1 to be made at the research level. In the motor/trophic category it is possible that he may additionally have suffered weakness and stiffness but the prior orthopedic intervention precluded consideration of these criteria.

HBO has been proposed as an effective treatment for decreasing pain and edema in patients with established CRPS of the wrist (17). Volunteers in this study received 15 HBO sessions on a once daily basis. Tissue hypoxia, as measured by skin capillary hemoglobin oxygenation (18) and skin lactate concentration (19) has been demonstrated in patients with CRPS.

A number of theoretical models have been suggested for the development of CRPS. These include 1) a focus on the afferent system such as the ischemic-reperfusion model of CRPS; 2) a primary central problem involving aberrant cortical reorganization; and 3) an efferent system problem involving a sympathetically mediated dysfunction.

Conclusion

Currently one of the most popular explanations suggests that there may be an initial peripheral insult such as an ischemic-reperfusion injury driving the process. HBO has thus been advanced as a plausible therapeutic strategy to minimize ischemic/reperfusion injury. This case report adds to the current literature by suggesting that CRPS Type 1 can still evolve despite ongoing HBO therapy.

Table 1. *Revised diagnostic categories for CRPS (16).*

	Sensory	Vasomotor	Sudomotor / Edema	Motor / Trophic
Symptoms	***Hyperesthesia***	***Temperature asymmetry***	***Edema***	Decreased range of movement
	Allodynia	***Skin color changes***	Sweating changes	Tremor
		Skin color asymmetry	Sweating asymmetry	Dystonia
				Weakness
				Nail or hair or ***skin changes***
Signs	***Hyperalgesia to pinprick***	Temperature asymmetry	***Edema***	Decreased range of movement
	Allodynia to light touch and/or to deep somatic pressure, and/or to joint movement	***Skin color changes***	Sweating changes	Tremor
		Skin color asymmetry	Sweating asymmetry	Dystonia
				Weakness
				Nail or hair or ***skin changes***

Bold = positive findings in this case.

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