

CASE REPORT

Fish Oil and BCQ™ as a Novel Treatment Approach to Primary Erythromelalgia: A Case Study

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Abstract

Erythromelalgia (EM) is a rare disorder, of which the pathophysiology is poorly understood. Though primary EM is thought to be an autosomal-dominant inherited disorder or of idiopathic origin, secondary EM is associated, most often, with myeloproliferative diseases. Pain management is the primary therapeutic target, however, individual patient responses vary widely, and no single treatment has been determined to be effective.

As such, EM is often associated with reduced quality of life and higher morbidity and mortality. This case study examines an 81-year-old Caucasian female patient with a diagnosis of primary EM, for which previous pharmaceutical therapies had proven ineffective. Therapeutic intervention included a novel, daily supplement regimen of fish oil and BCQ™.

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Introduction

Erythromelalgia (EM) is a rare neurovascular syndrome characterized by paroxysmal episodes of erythema, heat, tingling and burning pain in the extremities, which may be aggravated by exercise, stress and increased ambient air temperatures.¹⁻⁴ EM is more common in females than males and may develop at any age, however, is most frequently diagnosed in the fifth and sixth decades of life.³ The diagnosis of EM may be made clinically in the presence of five criteria, including burning extremity pain, pain aggravated by warming and relieved by cooling, and erythema and increased temperature of the affected skin.⁵ Ruling out myeloproliferative causes of EM is a top priority of clinical evaluation. Symptoms generally affect the feet, and occasionally the hands and face. Presentation is typically bilateral, although unilateral presentations have also been observed clinically. Patients with EM may seek relief from pain through extreme cooling methods including ice bath immersion, cold sock and glove application, and excessive use of fans. Complications related to hypothermia, frostbite, windburn and ulceration have been reported.^{3,6}

The etiology of primary EM is through autosomal-dominant inheritance or idiopathic origin, with the idiopathic subtype being most common. The genetic mutation associated with the inherited form of EM has not been documented in idiopathic primary EM and rarely in secondary cases.³

The pathophysiology of primary idiopathic EM is thought to be due to vascular and potentially neurologic abnormalities, resulting in a paradoxical presentation of hypoxia and hyperemia of the affected areas.^{1,7} Studies suggest that this is a result of pre-capillary sphincter constriction, with simultaneous opening of arteriovenous shunts, thereby increasing total perfusion of blood with reduced nutrient tissue perfusion. The resulting tissue hypoxia prompts increased local blood flow, which worsens symptoms of erythema, warmth and pain.^{7,8} Studies have shown that skin temperature of EM patients in the absence of a flare was lower than control subjects, which further suggests that vasoconstriction may play a role in the pathophysiology of this condition.⁷ It should also be noted that many patients with EM also have small fiber neuropathy, which might explain the effectiveness of certain treatments that address neuropathy in the management of EM.^{1,9}

EM is a rare condition of which there is no cure and limited data regarding gold standard of care, however pharmaceutical and non-pharmaceutical interventions aim to reduce pain and improve quality of life. Generally, a combination of topical and systemic pharmaceutical agents, as well as non-pharmacologic therapies are employed. Case reports have described use of topical