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Cutis marmorata telangiectatica congenital successfully treated with intense pulsed light therapy: A case report

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ABSTRACT

Cutis marmorata telangiectatica congenita is a rare disorder characterized by cutis marmorata, telangiectasia with or without recurrent ulcerations. It is a benign vascular anomaly with dilatation of capillaries and veins in the dermis. There is no satisfactory treatment for the ulcerative variety of cutis marmorata telangiectatica congenita. In this case, intense pulse light therapy was used with almost near total cure. Intense pulse light with its vascular filter of wavelength 550–1200 nm was used every fortnight till complete resolution of lesions.

ARTICLE HISTORY

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KEYWORDS

Cutis marmorata; intense pulse light; telangiectasia

Introduction

Cutis marmorata telangiectatica congenita (CMTC) is a rare congenital disorder characterized by cutis marmorata, telangiectasia with or without recurrent ulcerations and sometimes associated underlying cutaneous atrophy. Its incidence is sporadic, and its etiology is obscure. CMTC is a benign vascular anomaly with dilatation of capillaries and veins in the dermis. Usually a lower extremity is involved, but location on the trunk is not uncommon. A multitude of associated anomalies can occur, including limb asymmetry, hemangiomas and other vascular birthmarks, pigmented nevi, and aplasia cutis congenita. Diagnosis is usually evident on clinical examination (1). Bilateral involvement of breast in CMTC is reported by Lunge and Mahajan. (2) However, majority of patients have a good prognosis, with half demonstrating improvement of the mottled appearance over the first years.

Case report

A 20-year-old female patient with chief complaints of multiple, painful red-colored patches with multiple ulcerations over both the breasts since childhood. Pain was so severe that she could not even wear brassiere. Ulcerations used to heal with scarring followed by recurrence at other site.

Her health otherwise was absolutely normal. Dermatological examination revealed multiple, well-defined erythematous, reticulated plaques with few superficial ulcerations. There were few scars of healed ulcers and at places crusting was evident (Figure 1). Ulcers were tender with indurated base and covered with healthy granulation tissue. There were no regional or local lymphadenopathies. There was no abnormality in limbs, asymmetry of body, or any other vascular anomaly. There were no ocular, neurological, or alimentary canal changes. Her hemogram and biochemistry were also within normal limits. Collagen

vascular profile including ANA, dsDNA, and P-ANCA titers were within normal limits. Skin biopsy from the affected area showed classic dilated capillaries and veins in the dermis. Similar dilated capillaries and veins were seen in the subcutaneous tissue along with proliferation of vascular channels (Figures 2 and 3). The histological findings were suggestive of CMTC.

She has received multiple treatments before; in the form of systemic antibiotics, systemic steroids, pentoxifylline, oral niacinamide, and narrowband UVB with absolutely no benefit. Intense pulse light (IPL) (Forma TK, Israel; distributed in India by Geomedics Pharma Pvt Ltd.) therapy with 550–1200 nm vascular filter was started. To start with, 15 J/cm² energy was delivered in two subpulses of width 25 ms. Areas of ulcerations were skipped. The treatment was repeated every fortnight. She tolerated the treatment very well. Gradually energy was increased to 30 J/cm² keeping subpulses and pulse width constant. Patient was given short courses of azithromycin 500 mg once a day for 3 days as and when required to control the secondary infection in the ulcers.

After four sessions, there were no fresh ulcerations and old ulcers heal with superficial scarring. After six sessions, erythema and size of lesions reduced markedly [Figure 4(a)]. Patient also started wearing brassiere. After 8 sessions, there was about 90% improvement and after 10 sessions [Figure 4(b)] treatment was stopped and patient was followed up regularly for 1 year.

She never developed any ulcerations or pain or erythema during the 1-year follow-up period [Figure 4(c)].

Discussion

CMTC is a very rarely reported congenital vascular anomaly present at birth. Since the first case report by Von Lohuizen in 1922, more than 300 (3) cases have been reported worldwide to



Figure 1. Multiple reticulated hyperpigmented patches and ulcers on erythematous base on right breast.

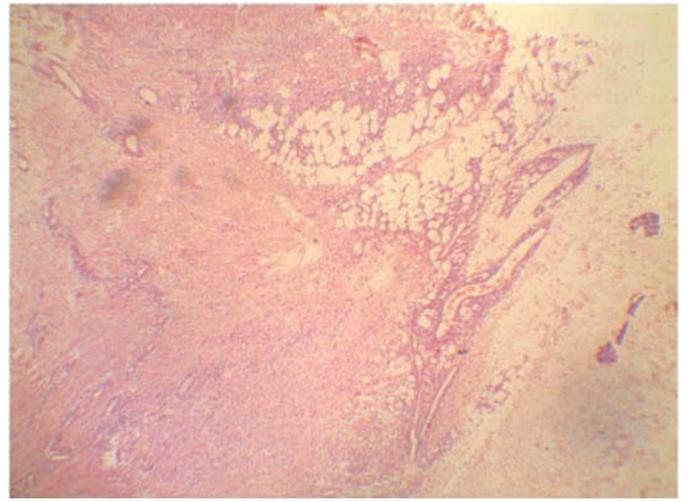


Figure 3. Dilated capillaries and veins in the subcutaneous tissue with proliferation of vascular channels.

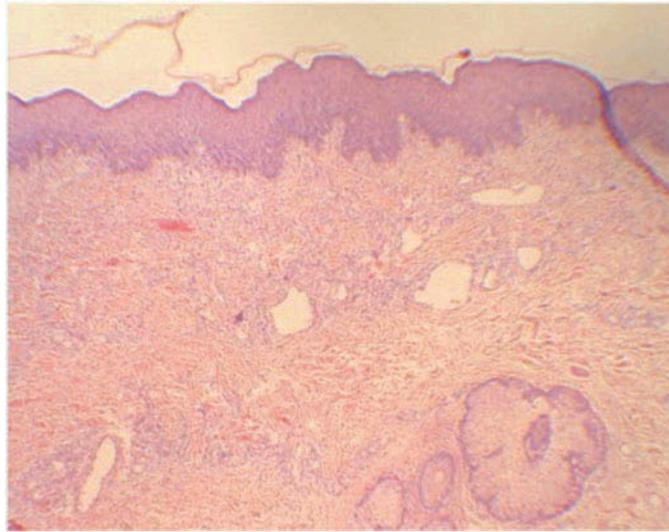


Figure 2. Dilated capillaries and veins in the dermis.

date. On reviewing the literature, it was found that there is no satisfactory treatment for this condition especially in ulcerative variety. Also cutis marmorata is a benign self-limiting condition

and does not require treatment but in ulcerative variety because of severe pain life becomes miserable for the patient. There are reports of phakomatosis pigmentovascularis type II-associated CMTC responding to combination laser treatment using Q-switched Alexandrite and long-pulsed dye laser (3). On review of literature on IPL therapy in vascular anomalies, we found that IPL reduces the inflammation by downregulating TNF- α (4). IPL enhances transforming growth factor beta1/smad3 signaling pathway (5), thereby inducing synthesis of dermal extracellular proteins *in vitro* (6). IPL also increases the content of dermal collagen and elastic fibers (7). All these factors help in reducing the inflammation as well as reduction of scarring, which was evident in our results of the therapy. IPL does help in correcting the dilatation of capillaries (8) by using vascular filter of wavelength 550–1200 nm. IPL therapy was started in this vascular anomaly because histologically it showed classical dilatation of capillaries in the dermis. IPL is also used in the erythematotelangiectatic type of rosacea (9,10,11) where it helps not only in reducing blood flow but also reduces the area of telangiectasia and thereby reducing erythema. It also helps in improving the skin texture. Schroeter and Neumann (12) and later on Angermeier (13) and Bjerring et al. (14) confirmed the use of IPL in facial telangiectasias. Clementoni et al (15) reported 87% of patients



Figure 4. (a) Reduction of lesion size with healing of ulcers after three sessions of IPL therapy. (b) Complete recovery with minimal scarring after 10 sessions of IPL therapy. (c) Follow up after 1 year no recurrence.

showed 75–100% clearance in facial telangiectasia. With the help of these reviews on IPL in the treatment of telangiectasia, this technology was used in treating CMTC with excellent result (Figure 4c).

It also revealed that the outcome of the treatment and the results were sustained for the 1-year follow-up period whereby there were no new ulcerations, erythema, and discomfort. IPL treatment definitely gives hope to the patient in the ulcerative variety of CMTC where rewarding result is seen in such a rare but miserable condition.

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Declaration of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the article.

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