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Case Report

Acquired Symmetrical Telangiectatic Macules of the Photo-Exposed Areas: Mastocytosis or Photodamage?

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Abstract

An uncommon form of macular telangiectatic eruption resembling cutaneous mastocytosis, previously known as Telangiectasia Macularis Eruptiva Perstans (TMEP), is described. The lesions have a very demarcated location, affecting only sun-exposed areas. We discuss the unusual clinical presentation in a 56-year-old woman on the light of the potential pathophysiologic events. Past clinical descriptions of TMPE had a common distribution in photo-exposed areas. It created some confusion with recently described forms of acquired telangiectatic disorders. In addition, the diagnosis of TMPE has been recently object of debate. In the end, TMEP has been removed from the present World Health Organization (WHO) classification of cutaneous mastocytosis. Some reports are still controversial about the differential diagnosis between photo dermatosis, acquired telangiectatic syndromes and skin limited cutaneous mastocytosis, which may share some similar histological findings.

Keywords: Telangiectasia Macularis Eruptiva Perstans; pathophysiologic Events; Telangiectatic Syndromes

Introduction

Telangiectasias are chronic dilation of capillaries and venules that create linear or aracneiform shapes visible on the surface of the skin. The color ranges from brilliant red to the shades of blu with regard to the oxygenation of the blood and the dept of the vessels. They may be a typical sign of a few primary (mainly hereditary) disorders, but they are commonly secondary to several conditions [1].

A challenging problem in the diagnosis of acquired telangiectasias is the differentiation between acquired telangiectatic macules, frequently localized in sun-exposed areas and a clinical subtype of cutaneous mastocytosis previously known as Telangiectasia Macularis Eruptiva Perstans (TMEP) [2,3].

Cutaneous mastocytosis (CM) can be associated to systemic complication with the involvement of internal organs. The skin lesions are usually several and develop progressively associated with variable intensity of itching. Pediatric mastocytosis is well-known (mainly solitary mastocitoma of the skin). Systemic Mastocytosis (SM) is uncommon, but it is sometimes a severe condition which should be treated as early as possible [4].

Cutaneous mastocytosis has been classically described in 4 main variants: cutaneous mastocytoma, urticaria pigmentosa, cutaneous diffuse mastocytosis and telangiectasia macularis eruptiva perstans (Table 1) [5]. They are mainly limited to the skin, but the presence of systemic organic involvement is always possible as potential complication.

Hepatosplenomegaly, bone involvement and multiple lymphadenopathies are the most common signs of SM. TMEP is uncommon. In 2016, an international task force of different specialties experts proposed a new classification of cutaneous mastocytosis that excluded the term TMEP [6-8]. It was considered to overlap with urticaria pigmentosa. However, various clinical reports supported the concept of photodamaging as the main factor producing the clinical presentation of some cases of previously suspected TMEP [3].

Maculopapular Cutaneous Mastocytosis (MPCM)/urticaria pigmentosa
Diffuse Cutaneous Mastocytosis (DCM)
Solitary mastocytoma of skin

Table 1: WHO Classification of cutaneous mastocytosis 2016.

Case Report

A 56-year-old woman presented with asymptomatic cutaneous telangiectatic lesions. She declared that the lesions developed without any known trigger factor and had a progressive evolution since she was 46. They were totally asymptomatic. The first areas involved were her upper arms and chest and over the following years extended to her abdomen, back and lower extremities (Fig. 1-3). She did not complain any clinical systemic condition. A periodic control of blood exams, including homocysteinemia was performed. On examination, she showed the presence of diffuse teleangiectasias of the facial and trunk skin ranging from 0,5 to 4 cm of diameter. They were concentrated in the upper arms, chest, face, shoulders and back. Fewer lesions were present on abdomen and lower limbs. The lesions were symmetrically distributed and essentially concentrated on sun-exposed areas, with evident sparing of the skin usually covered by the clothes and a clear demarcation line particularly evident on shoulders and chest. The Darier sign was negative [4,5].

Hepatosplenomegaly or palpable lymph nodes were not found clinically and ecographycally. The laboratory tests included complete blood count, peripheral blood film examination and liver function tests. They were all within normal limits except for and increase in white blood cells (WBC 15640/ul, N 8430/ul, L 6170/ul) and ANA 1:320 (nucleolar pattern). Ant-nDNA and anti-ENA antibodies were not present. Tryptase test resulted in a normal range. The histological examination of a 4 mm punch-biopsy of the lesional skin showed dilated vessels in the papillary dermis and scant perivascular infiltrates of mast-cells and lymphocytes (Fig. 4). Numerous mast cells (CD117+) were present around superficial vessels (Fig. 5). A diagnosis of cutaneous mastocytosis, previously known as telangiectasia macularis eruptiva perstans, no longer classified as a separate variant (WHO 2022), was made on the basis of the clinical presentation and the typical histopathological findings.

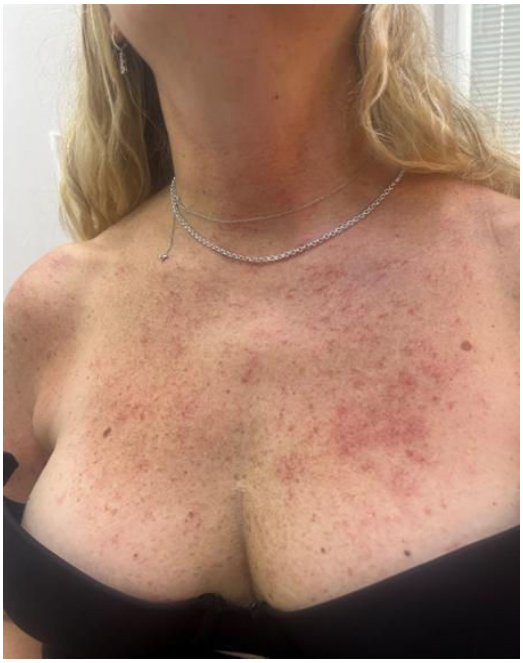
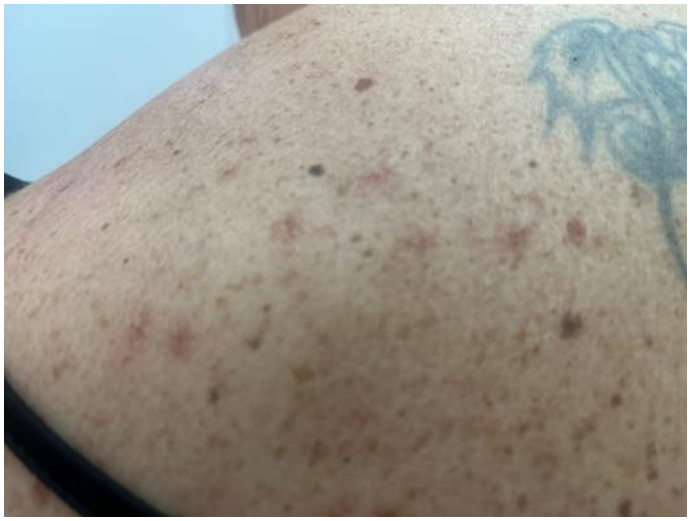


Figure 1:



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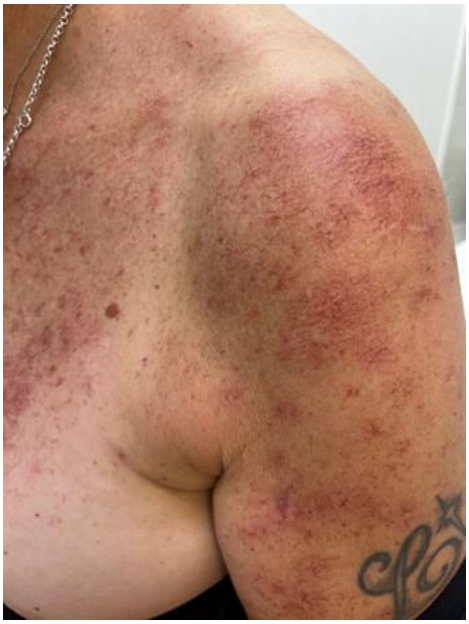

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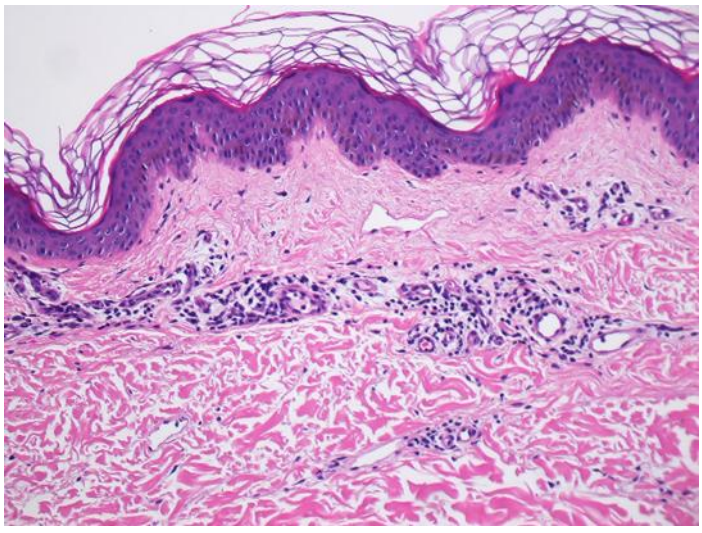

Figure 4:

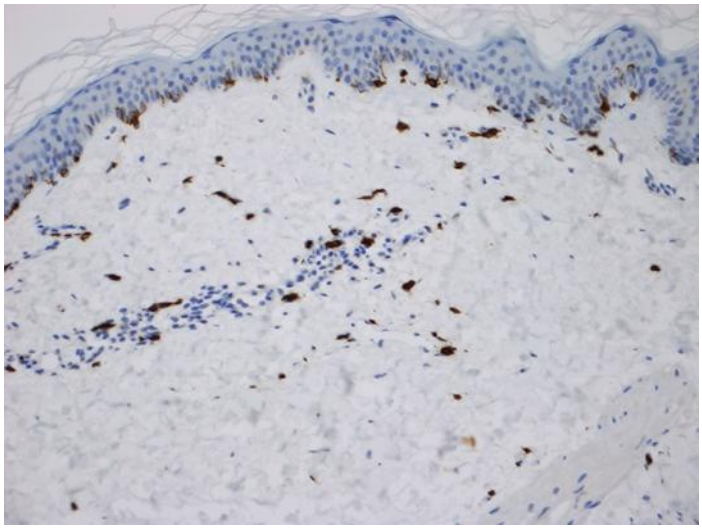

Figure 5:

Discussion

Parkes Weber in 1930 described, with the term TMEP, an uncommon clinical condition of mastocytosis. Several cases were reported later. The most recent classification of cutaneous mastocytosis, which excluded the term of TMEP, created confusion among clinicians and researchers who have some concerns in the identification of very similar clinical disorders.

Telangiectatic clinical presentations of cutaneous mastocytosis no longer classified as a separate variant. A classical cutaneous marker of mastocytosis is the Darier sign which consists in a quick development of an area of redness and swelling within a few minutes after scratching of the lesion. Systemic involvement is possible. It may involve Bone Marrow (BM), gastrointestinal tract, liver, spleen and lymph nodes. Multiple myeloma and polycythemia vera have been reported in association with cutaneous mastocytosis. Tryptase is a vasoactive immunoregulatory mediator found in mast cell granules.

Increased serum total tryptase levels >20 ng/ml should alert as a sign of SM owing to its elevation in most patients with SM. In most cases of cutaneous mastocytosis a consistent number of mast cells are typically found surrounding venules and dilated capillaries in the dermis.

In patients with clinical signs and symptoms of cutaneous mastocytosis confirmed by histology, WHO criteria for SM include screening for serum tryptase level, BM histology (tryptase and/or KIT (CD117) immunostaining), mast cells immunophenotyping (CD25 and CD2 expression), KITD816V mutation screening in the BM, blood or extracutaneous specimen and FIP1L1-PDGFRA screening in the BM and blood if eosinophilia is present. If systemic involvement is not found, follow up should be done with a blood count, serum tryptase level and observation [9]. The treatment is firstly symptomatic. No gold standard treatment to date is reported. Moon and coworkers have recently hypothesized that an increased number of mast cells dilated vessels in the dermis and the abnormal presence of mast-cells around them might be a photoaging-related reactive process of chronic sun-exposure, which consequently leads to the formation of characteristic telangiectatic hyper pigmentary macules through melanogenic mediators, rather than a subtype of cutaneous mastocytosis [3]. This hypothesis has not been confirmed yet [10].

Conclusion

The concerns we had for our patient are very similar to the case reported by Moon and coworkers. The presence of not-significant number of mast-cells around the dilated capillaries in the dermis creates always doubts in the clinician. If symptoms are not present and a systemic involvement may be reasonably excluded, the follow up of the patients remains fundamental. A 1 year follow up in the patient reported did not show any clinical change. She regularly applied body moisturizing cream and photoprotector products.

Conflict of Interest

The authors declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

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Data Availability Statement
Not applicable.

Ethical Statement
The project did not meet the definition of human subject research under the purview of the IRB according to federal regulations and therefore, was exempt.

Informed Consent Statement
Informed consent was taken for this study.

Authors' Contributions
All authors contributed equally to this paper.

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