Case for diagnosis

Caso para diagnóstico

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CASE REPORT

A 27 year-old-man with no known history of personal or familial illnesses was referred to our clinic with a 10 year history of cervical telangiectasias with no associated symptoms. The patient denied any preceding or simultaneous episodes of disease with this dermatosis, as well as concomitant or recent use of any medication. We found no changes on his physical exam, except the presence of groups of blancheable, erythematous macules, of a bright red tone, with a Blaschko lines distribution and localized only on the right lateral aspect of the neck (Figures 1 and 2). Blood work did not reveal any changes, namely in liver enzymes or infectious serologies. What is your diagnosis?

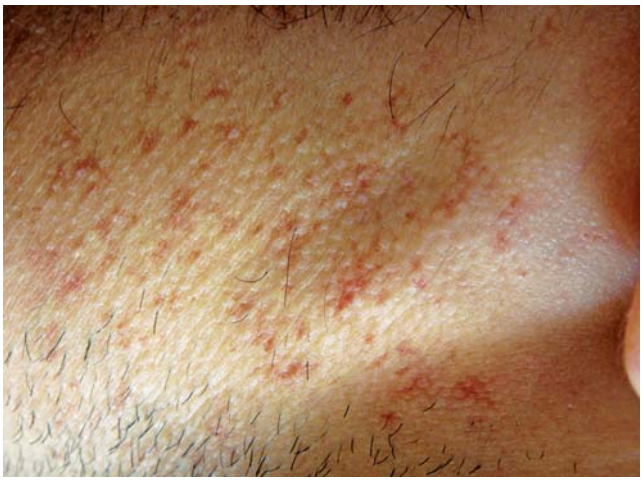

FIGURE 2: Detail of telangiectasias shown on figure 1

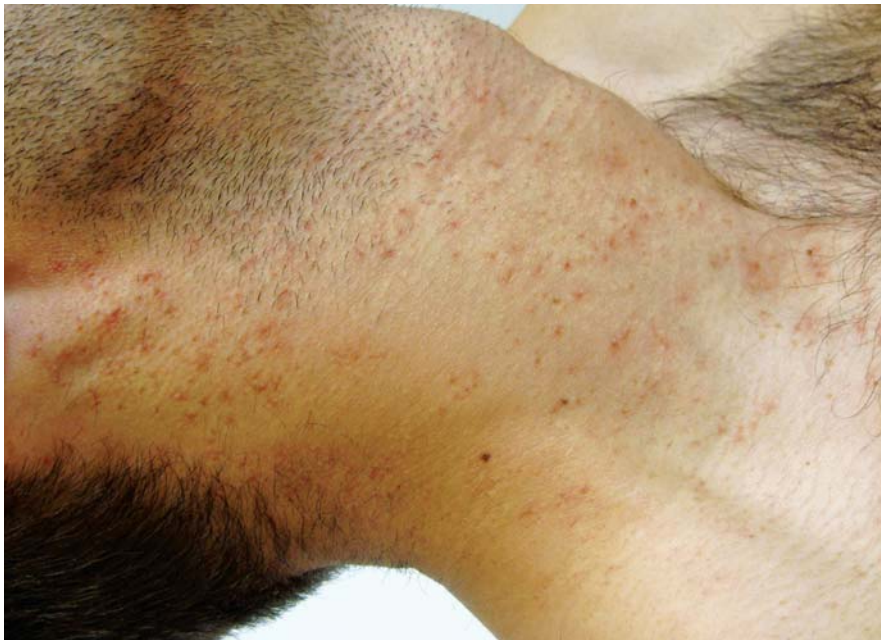

FIGURE 1: Telangiectasias on the right lateral aspect of the neck

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DISCUSSION

Unilateral Nevoid Telangiectasia (UNT) was initially described by Blaschko in 1899.[1] Its incidence is unknown, although it's probably underreported and more frequent than previously recognized.[2]

UNT is a dermal vascular malformation of unknown cause and two presentations, congenital or acquired.[3] There is controversy in regard to the existence of a somatic mutation during embryogenesis in the congenital form or, in the acquired form, the elevation of estrogen levels or vasoactive substances that may, in theory, be revealing of a state of hidden mosaicism.[4-6] Only one publication to date has identified the elevation of estrogen receptors in lesional skin.[7] Others have proposed the role of a still unidentified vasoactive substance.[8]

However, UNT favors female individuals, around puberty, that are on hormonal contraception, pregnant women, or individuals with chronic liver disease.[5] Indolent and asymptomatic, it is characterized by groups of telangiectasias in variable number and sizes, with a linear, dermatomal, multidermatomal or Blaschko lines distribution, the 3rd - 4th cervical dermatomes, face, shoulder, arm and upper trunk being the most commonly involved locations.[2,4] Among main differential diagnosis are: 1) primary telangiectasias, mainly Linear Atrophoderma of Moulin, telangiectatic variant, or Angioma Serpiginosum; and 2) secondary telangiectasias, like Erithema ab Igne or use of topical corticosteroids.[2]

Pregnancy tests, dosing of liver enzymes or serologies for hepatotropic viruses may be used.[2,4,5] Histologically, the unspecific identification of capillary dilations on the superficial dermis only confirms the clinical diagnosis of telangiectasias and may require biopsy of peri-lesional skin for comparison.[3] Laser-Doppler Fluxometry allows the noninvasive identification of local blood flow alterations.[9] In the case of hyperestrogenemia related with ectopical estrogens, their removal may improve the disease.[4] The remaining cases may be approached cosmetically with the use of camouflage, Nd:YAG laser or Pulse Dye Laser.

The identification of this case of acquired idiopathic UNT in a healthy male individual may not be as rare as previously thought, a dermatosis to be equated in the more frequent differential diagnoses of telangiectasias. ❑

Abstract: A 27 year-old-man, with no known personal or familial history of disease, mentioned a 10-year history of asymptomatic groups of telangiectasias, with a Blaschko lines distribution on the right lateral aspect of the neck and asymptomatic. He denied any episodes of disease or drug intake that could be associated with the disease. Blood work had no changes, namely of liver enzymes or infectious serologies. The clinical diagnosis of Idiopathic Acquired Unilateral Nevoid Telangiectasia was made, an uncommon, benign vascular malformation. The patient declined doing a cutaneous biopsy or treatment with a cosmetic intent.
Keywords: Estrogens; Mosaicism; Telangiectasis

Resumo: Um homem de 27 anos, sem quaisquer antecedentes patológicos pessoais ou familiares conhecidos, mencionava dermatose com cerca de 10 anos de evolução, caracterizada por agrupamentos de telangiectasias, de distribuição blaschkóide, na face lateral direita do pescoço e assintomáticas. Negava quaisquer episódios de doença ou toma de fármacos que pudessem estar relacionados com esta doença. As análises sanguíneas do doente estavam inalteradas, nomeadamente os enzimas hepáticos e serologias infecciosas. Foi efectuado o diagnóstico clínico de Telangiectasia Nevóide Unilateral Adquirida, idiopática, uma malformação vascular benigna pouco comum. O doente prescindiu da realização de biópsia cutânea ou tratamento de intenção cosmética.
Palavras-chave: Estrogênios; Mosaicismo; Telangiectasia

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