

Spontaneous extracranial hemorrhagic phenomena, autoerythrocyte sensitization syndrome and anticardiolipin antibodies: Is there any relationship?

Sir,

We read with interest the letter of Gupta *et al.*, in which two cases of recurrent forehead ecchymoses with headache were reported.¹ Spontaneous extracranial hemorrhagic phenomena is an infrequent and poorly understood feature of primary headache disorders such as migraine, tension-type headaches and trigeminal autonomic cephalalgias, characterized by spontaneous ecchymosis ipsilateral to the side of the headache in 2/3 of the cases and bilateral in the rest. The ecchymosis resolves in 2–10 days. The coagulation studies are always normal and skin biopsies show extravasation of red blood cells in the dermis.

The autoerythrocyte sensitization syndrome (Gardner-Diamond syndrome) is an extremely rare disorder characterized by spontaneous bruises preferably affecting women with psychoemotional disorders (not an imperative requisite for diagnosis) or with antecedent of physical trauma. It has been suggested that these patients auto-sensitize to their own blood, specifically to phosphatidylserine which is part of the membrane of red blood cells.² Skin biopsy of the bruises shows extravasation of erythrocytes in at least 62% of cases in some reviews^{3,4} and the presence of headache in 13%.⁴

Based on phosphatidylserine is a phospholipid, positive anticardiolipin antibodies have been described in this condition.⁵ However, the exact relationship between these antibodies and Gardner-Diamond syndrome is not clear.

Anticardiolipin antibodies and antiphosphatidylserine/prothrombin complex have been described also in a possible forme fruste of antiphospholipid syndrome.⁶ As anticardiolipin antibodies are related with antiphospholipid syndrome which is characterized by the occurrence of venous and arterial thrombosis rather than spontaneous bruises, among other signs, we hypothesize that perhaps spontaneous extracranial hemorrhagic phenomena could

be a form of Gardner-Diamond syndrome, and we suggest that anticardiolipin antibodies should be measured in those cases.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Sudy E, Urbina F. Spontaneous extracranial hemorrhagic phenomena, autoerythrocyte sensitization syndrome and anticardiolipin antibodies: Is there any relationship? *Indian J Dermatol Venereol Leprol* 2021;87:380.

Received: May, 2020 Accepted: January, 2021 Published: April, 2021

DOI: 10.25259/IJDVL_621_20 PMID: 33871187

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