

CASE REPORT

Six Cases of Nevus Oligemicus

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Abstract. Hamartoma or nevus oligemicus is an uncommon lesion that is characterized by selective vasoconstriction of the deep dermal vascular plexus with respect to the superficial one and whose cause has not been clearly established. This selective vasoconstriction gives rise to fixed, acquired, and asymptomatic lesions in the form of livid, erythematous macules that are typically cold to touch compared with surrounding skin.

We report the cases of 6 young men with lesions clinically compatible with nevus oligemicus on the abdomen and flanks. Measurement of the surface temperature of the lesion revealed a decrease of up to 2.5°C with respect to healthy surrounding skin and allowed a definitive diagnosis to be made.

We describe the additional studies undertaken, the differential diagnosis, and the possible etiologic agents, and discuss the cases reported in the literature to date.

In our opinion, nevus oligemicus is an underdiagnosed lesion that is much more common than has been reported in the literature.

Key words: hamartoma, nevus, oligemicus.

NEVUS OLIGEMICUS. APORTACIÓN DE SEIS CASOS

Resumen. El hamartoma o *nevus oligemicus* es una entidad infrecuente, de etiopatogenia poco aclarada, en la que se produce una vasoconstricción selectiva del plexo vascular profundo dérmico con relación al superficial. Estas alteraciones funcionales dan lugar a lesiones fijas, adquiridas y asintomáticas a modo de máculas lívidas, eritematosas, típicamente frías al tacto si se compara con la piel sana adyacente.

Comunicamos los casos de 6 varones jóvenes con lesiones en el abdomen y en los flancos, compatibles clínicamente con *nevus oligemicus*. La medición de la temperatura superficial de la lesión, constatando un descenso de hasta 2,5 °C respecto de la piel sana adyacente, permitió realizar el diagnóstico definitivo.

Se describen los estudios complementarios realizados, los diagnósticos diferenciales, los posibles agentes etiológicos responsables y se comentan los casos referidos en la literatura hasta el momento.

Consideramos que el *nevus oligemicus* es una entidad infradiagnosticada y mucho más frecuente de lo referido en la literatura.

Palabras clave: hamartoma, nevus, *oligemicus*.

Introduction

The etymology of *oligemicus* is *oligo*, meaning little, and *emicus*, meaning blood; thus, the term refers to “poor vascularization.” Hamartoma or nevus anemicus is defined as a circumscribed cutaneous abnormality characterized by localized vasoconstriction of the deep dermal vascular plexus (which regulates temperature) compared with more

superficial dermal vessels (which handle nutrition).¹ Clinically, the condition leads to cold, erythematous lesions. Localized hypothermia is a key finding for the diagnosis of the condition.

Nevus oligemicus was first described in 1981² and only an additional 3 cases have been reported. We report another 6 cases diagnosed in the past 5 years.

Case Description

The clinical and epidemiologic findings of the patients are summarized in Table 1. The patients were 6 men between 26 and 57 years of age; all were considerably overweight and had a sedentary profession (4 truck drivers, 1 candidate

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Table 1. Clinical and Epidemiologic Findings

	Profession	Sex/Age, y	Precipitating Factors	Clinical Findings
Patient 1	Truck driver	M/4	None	Cold erythema
Patient 2	Truck driver	M/43	None	Cold, livid erythema
Patient 3	Candidate for government position	M/26	None	Cold erythema
Patient 4	Truck driver	M/49	None	Cold erythema
Patient 5	Truck driver	M/54	None	Cold erythema
Patient 6	Bank employee	M/57	None	Cold erythema

Abbreviations: M, male; ND, not done.

Table 2. Clinical and Epidemiologic Findings Previously Described in the Literature

Cases	Profession	Sex/Age, y	Precipitating Factors	Clinical Findings/Course
Fays et al ¹	NK	M/46	None	Livid erythema
Davies et al ²	NK	M/16	Cold baths	Livid area
Friedel et al ³	NK	F/15	None	Livid, cyanotic area
Dupré et al ⁴	NK	M/45	None	Cold, cyanotic, erythematous area

Abbreviations: F, female; M, male; ND, not done; NK, not known.



Figure 1. Patient 2. Striae distensae and erythematous-cyanotic plaque following 15-kg weight loss.



Figure 2. Patient 6. Cold erythema on the flanks.

for a government position, and 1 bank employee). Patient 4 had undergone appendectomy and surgery for melanoma on the back 6 years earlier. All others had no relevant personal or family history. In 5 of the patients, the dermatologic examination revealed a fixed erythematous cyanotic macule with irregular borders in the middle of the abdomen (Figure 1). In the sixth, the erythematous area appeared on both flanks (Figure 2). The lesions remained stable and whitened on pressure. The skin was cold in the

erythematous area, particularly in the middle of the plaque.

The laboratory workup included complete blood count, complete biochemistry, antinuclear antibodies, lupus anticoagulant, cryoglobulins, and *Borrelia burgdorferi* serology, and all results were normal or negative.

A skin biopsy was taken in the first 3 cases. The histologic study revealed a normal number of dermal vessels, although with dilatation of the papillary dermal capillaries and endothelial hyperplasia. In the reticular dermis, hyperplasia

Site	Course	Vasodilators	Vasoconstrictors	Axon Reflex
Abdomen	Stable	Thurfyl nicotinate +	0.05% Clobetasol propionate +	Histamine +
Abdomen	Stable	Thurfyl nicotinate +	0.05% Clobetasol propionate +	Histamine +
Flank	Stable	Thurfyl nicotinate +	0.05% Clobetasol propionate +	Histamine +
Abdomen	Stable	Thurfyl nicotinate +	0.05% Clobetasol propionate +	Histamine +
Abdomen	Stable	Thurfyl nicotinate +	0.05% Clobetasol propionate +	Histamine +
Abdomen	Stable	Thurfyl nicotinate +	0.05% Clobetasol propionate +	ND

Site	Course	Vasodilators	Vasoconstrictors	Axon Reflex
Flank	Stable	Thurfyl nicotinate +	Scopolamine + Norepinephrine + 0.05% Clobetasol propionate +	Histamine +
Hand	Stable	Phentolamine +	Propranolol + Adrenaline +	Histamine +
Chest and thigh	Stable	Thurfyl nicotinate +	Epinephrine + Betamethasone 0.05% +	Histamine +
Abdomen and thighs	Stable	ND	ND	ND

of the endothelial cells had caused obstruction of the capillary lumens (Figure 3).

Analysis of surface skin temperature (with an MX-TDI2388 contact thermometer, MX onda, Madrid, Spain) showed a decrease of 2°C to 2.5°C in the affected area compared with adjacent healthy skin.

Various pharmacological tests with histamine, thurfyl nicotinate, and 0.05% clobetasol propionate showed normal vasomotor responses of both the healthy skin and the hamartoma. An intradermal injection of histamine (1.0 ng histamine in 0.1 mL saline solution) plus topical application of thurfyl nicotinate caused a bump in the first case and erythema and edema in both healthy and affected skin in the second. The application of 0.05% clobetasol propionate ointment with an occlusive dressing for 2 hours caused identical intensity in the whitening of both areas.

The clinical characteristics and decreased temperature of affected skin led to a diagnosis of nevus anemicus. Only the first case received treatment with 20 mg of oral prednisone for 1 month, with a subsequent decrease to 10 mg daily the second month, with no changes observed. None of the other patients received any medications.

No improvement or worsening of the original lesions and no new lesions were observed during the follow-up period, which varied from 2 to 6 years. Patient 5 lost 15 kg, but with no changes in the plaque (Figure 1).

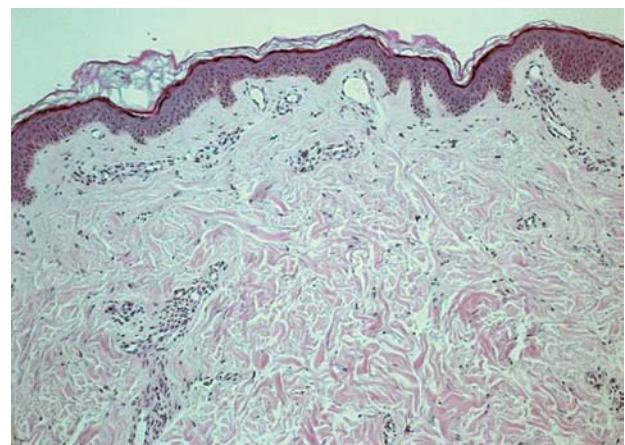


Figure 3. Histologic findings of livid erythema, with dilated vessels in the papillary dermis. Hematoxylin-eosin, ×40.

Discussion

Hamartoma or nevus anemicus is a common entity, understood as a functional, rather than anatomical, abnormality.³ The etiology is not well known, but may be due to hormone receptor abnormalities, with variations in adrenergic activity,¹ increased sympathetic tone, and

consequently vasoconstriction and slower deep dermal vascular flow,³ thus cooling the lesion surface. However, the superficial vascular plexus (responsible for nutrition) is intact or thought to have suffered relative vasodilation, which led in our patients to the erythematous appearance of the lesions.^{2,4}

These 6 cases are consistent with the diagnosis of nevus oligemicus, first described by Davies et al² in 1981 in a man with an area of fixed, persistent, livid erythema on the trunk, with a striking drop in local temperature compared with adjacent healthy skin.

Clinically, the lesions present as erythematous-violaceous areas that are fixed, irregular, and cyanotic or, in some cases, as whitish, telangiectatic plaques.¹ They are usually located in the abdominal region and thighs, but cases have also been described on the hands⁴ or chest and thighs.⁵ The main clinical characteristic is the persistence of cold skin temperatures to touch, in the absence of evident arterial ischemia in the vicinity.^{1,2,4,5} The main diagnostic sign is skin temperature, which is usually at least 2°C lower than the adjacent skin.^{1,2,4,5}

No triggers have been described except for cold baths, as reported in 1982 by Dupré and Viraben⁴ (Table 2). We found possible factors favoring this condition among the patients described in this article because all were obese and had a sedentary lifestyle related to their profession. Although 1 patient lost 15 kg, resolution of the lesions did not occur. Despite most lesions appearing above the belt adjustment area, we were unable to determine if pressure was the source of the abnormality because the nevi oligemicus did not disappear when a belt was not worn.

Laboratory analysis and immunologic studies are normal and histopathology shows discrete vasodilatation of the superficial dermis and obstruction of the deep vessels, as reported by Dupré and Viraben⁴ and observed

in the 3 patients from our study in whom biopsy was performed. However, skin biopsy is not essential to diagnose the condition, which is essentially clinical and demonstrated by temperature measurement and localized hypothermia.

The differential diagnosis can be performed with inflammatory erythema and capillary hemangioma.² In the first case, the erythema is not fixed, and there are local increases in temperature and epidermal changes seen in the histologic study. Hemangiomas are associated with characteristic histologic findings not found in nevus oligemicus.

We consider nevus oligemicus to be a common condition, perhaps underreported by patients, underdiagnosed, or even minimally reported in dermatology journals. We contribute 6 new cases to add to those reported in the literature.

Conflicts of Interest

The authors declare no conflicts of interest.

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