CASOS PARA EL DIAGNÓSTICO

[Translated article] Erythematous nodule on the upper lip
Nódulo eritematoso en labio superior

Clinical history
A 37-year-old man, with no personal or family history of interest presented with a 2-month history of asymptomatic lesions on his upper lip. He denied the occurrence of any hemorrhages and had not seen any ulcers in the lesions.

Physical examination revealed the presence of an erythematous nodule of a smooth surface and semi-solid consistency on the upper lip (fig. 1A), along with an additional asymptomatic papular lesion on the same mucosa (fig. 1B). Dermoscopy (fig. 1C) revealed the presence of a homogeneous red pattern with whitish areas surrounded by dilated tortuous vessels, some resembling a forked vascular pattern.

Histopathological examination of a 4 mm punch biopsy confirmed the presence of an intradermal tumor lesion composed of a vascular proliferation of prominent endothelium, along with a predominantly lymphocytic inflammatory infiltrate with few interspersed eosinophils.

No additional tests were performed for diagnostic purposes.

What is the diagnosis?

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Diagnosis and comments

The diagnosis achieved was angiolymphoid hyperplasia with eosinophilia (ALHE).

Disease progression and treatment

Although the patient was informed about the possibility of spontaneous lesion resolution, he insisted on being treated, and received 2 sessions (595 nm, 7 mm diameter, 7 J/cm² fluence, and 0.5 ms pulses; 2 pulses per lesion; air cooling) of pulsed dye laser (PDL), achieving complete resolution with an almost perfect aesthetic appearance (fig. 2A-C). The skin alterations did not relapse 6 months into treatment.

Comments

ALHE is a subtype of epithelioid hemangiendothelioma, within benign angiolymphoproliferative processes, which is associated with a greater inflammatory component. It is a rare entity more prevalent in middle-aged women characterized by the appearance of erythematous-violaceous papules and nodules and is usually located on the head and neck. Lip or oral mucosa involvement is a rare finding, as it predominantly affects the periauricular area (36.3%), face (28.2%), and scalp (17.3%). Although these papules and nodules often remain asymptomatic, they may be associated with pruritus. Additionally, although their etiopathogenesis is unknown to this date, they could be due to reactive vascular proliferation in response to infections, hormonal factors, or trauma. The occurrence of ALHE after getting a tattoo done supports this latter hypothesis. Histological examination shows the proliferation of vascular structures of varying calibers with prominent epithelioid endothelial cells, along with a lymphocytic infiltrate and the presence of eosinophils.

Differential diagnosis includes entities such as cutaneous angiomatoid nodule (CEAN), cutaneous sarcoïdosis, pyogenic granuloma, cutaneous lymphoma, Kaposi’s sarcoma, and various types of epithelioid neoplasms. Among these, ALHE should be distinguished from other vascular epithelioid lesions such as epithelioid hemangiendothelioma, pseudomalignant hemangiendothelioma, and epithelioid angiosarcoma (EA).

Histopathological characteristics and the use of an immunohistochemical panel are used to distinguish one entity from the other. Although ALHE generally expresses the switch regulatory protein 3 (SPP3), conditions such as pseudomalignant hemangiendothelioma can also express it. Superficial location, good delimitation, and the absence of an infiltrative pattern with a low degree of nuclear atypia and mitotic figures, and negativity for podoplanin and avian myelocytomatosis viral oncogene homolog, distinguish ALHE from EA.

The management of ALHE is not standardized. Excision is usually performed to treat single lesions, while therapies with variable results include topical and intralesional corticosteroids, cryoagulation, pentoxifylline, or various laser modalities are used to treat multiple lesions. In published cases, PDL has turned out to be a safe therapy that improves symptoms and the aesthetic appearance of lesions, even leading to their disappearance, as it happened with our case. The mechanism of action of PDL induces photothermolysis, which eventually leads to the selective destruction of vascular lesions, thus reducing damage to surrounding structures, and facilitating faster healing compared with other therapies such as carbon dioxide. This has turned PDL into an attractive option for the management of ALHE since this is a benign condition that tends to recur frequently despite previous therapies.

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Conflicts of interest

None declared.

References


M.D. Pegalajar-García, I. Pérez-López, F.J. Navarro-Triviño* Q2

Servicio de Dermatología, Hospital Universitario San Cecilio, Granada, Spain

*Corresponding author.

E-mail address: fnntmed@gmail.com (F.J. Navarro-Triviño).