

CASE REPORTS

A Histopathologic and Imaging Study of Verrucous Hemangioma

A.A. Garrido-Ríos, L. Sánchez-Velicia, J.M. Marino-Harrison, M.V. Torrero-Antón, and A. Miranda-Romero

Servicio de Dermatología, Hospital Clínico Universitario de Valladolid, Facultad de Medicina de Valladolid, Valladolid, Spain

Abstract. The terms verrucous hemangioma and angiokeratoma circumscriptum have been used interchangeably in the literature to define clinically similar lesions. From a histologic perspective, however, angiokeratoma circumscriptum is limited to the papillary dermis whereas verrucous hemangioma extends as far as the hypodermis. We describe the case of a 38-year-old woman who consulted for a lesion on the right thigh that was initially diagnosed as angiokeratoma; magnetic resonance imaging, however, led to a final diagnosis of verrucous hemangioma.

Key words: angiokeratoma circumscriptum, verrucous hemangioma.

HEMANGIOMA VERRUGOSO. ESTUDIO HISTOPATOLÓGICO Y RADIOLÓGICO

Resumen. Los términos hemangioma verrugoso y angioqueratoma circunscrito han sido utilizados en la literatura de manera indistinta para definir lesiones clínicamente similares; sin embargo, desde un punto de vista histológico, el angioqueratoma circunscrito se limita a la dermis papilar, mientras que el hemangioma verrugoso se extiende hasta la hipodermis. Presentamos el caso de una mujer de 38 años que consultó por una lesión en el muslo derecho que fue inicialmente diagnosticada de angioqueratoma; sin embargo, tras la realización de una resonancia magnética nuclear se llegó a un diagnóstico final de hemangioma verrugoso.

Palabras clave: angioqueratoma circunscrito, hemangioma verrugoso.

Introduction

Verrucous hemangioma and angiokeratoma are terms that have been used indistinctly in the literature to refer to clinically similar vascular lesions. However, it is important to differentiate between these 2 conditions in order to institute a correct approach to management.

We present the case of a patient who underwent excision of a verrucous hemangioma on the right flank in 2001, and who was subsequently seen for a lesion on the right lower limb.

Case Description

The patient was a 38-year-old woman with no known drug allergies. In 2001, she underwent excision of a verrucous hemangioma on the right flank.

She came to the dermatology department 5 years later because she was concerned about a lesion that had been present on the right thigh since childhood but that had grown progressively in recent months and was occasionally itchy.

On physical examination, there were several round, well-defined, reddish papules that tended to group together, situated on the external aspect of the upper third of the right thigh (Figure 1).

Round, skin-colored or slightly reddish papules with a verrucous surface and measuring 3 to 4 mm in diameter were observed on the right flank around the scar where the verrucous hemangioma had been excised 5 years earlier (Figure 2).

There was no asymmetry of the lower limbs and the lesions were not pulsatile.

An incisional biopsy was taken from the lesion on the right thigh, revealing irregular acanthosis of the epidermis, elongation of the rete ridges, orthokeratosis with foci of parakeratosis, and hyperpigmentation of the basal layer. The blood vessels of the papillary dermis were dilated and cavernous. At the papillary border, there was a tendency of the epidermis to surround the dermal vascular dilatations (Figure 3). The lesion was CD34 positive.

The clinical and histological findings were compatible with the diagnosis of angiokeratoma circumscriptum.

Correspondence:
Anastasia A. Garrido Ríos
C/ Huertas, 3, 4^º P
47005 Valladolid, Spain
natachagarrido@hotmail.com

In addition, histology of the verrucous hemangioma excised 5 years earlier revealed a slightly hyperplastic epidermis with papillomatosis, hyperkeratosis, and acanthosis, and large, dilated vascular structures lined by a fine layer of endothelial cells with no atypia were seen in

the superficial dermis (Figure 4). These dilatations contained a large quantity of red blood cells. Another vascular proliferation formed of large, dilated vessels of venous appearance could be seen in the deep dermis and in the subcutaneous cellular tissue; these vessels contained hematic material and were surrounded by a proliferation of small capillaries. This lesion was also positive for CD34.

We were therefore faced with 2 lesions with very similar findings in the epidermis (acanthosis, papillomatosis, and hyperkeratosis) and superficial dermis (dilated blood vessels) and both of which expressed CD34. However, the depth of infiltration of the lesion on the thigh could not be assessed because the biopsy did not include subcutaneous tissue. As the patient refused to undergo further biopsy, magnetic resonance imaging was performed to visualize the deep part of the lesion. This revealed tortuous, tubular images of reticular morphology penetrating the subcutaneous cellular tissue and making contact with but not penetrating the underlying muscles, visible from the level of L1-L2 down to the upper third of the thigh (Figure 5).

Based on the clinical, histopathologic, and imaging study findings, we diagnosed the lesion on the thigh as a continuation of the verrucous hemangioma previously excised from the right flank.

Figure 1. Round, reddish papules grouped on the right thigh.



Figure 2. Lesion on the right thigh and papules around the scar on the right flank.



Discussion

Verrucous hemangioma was first described by Halter in 1937, but it was Imperial and Helwig¹ who, in 1967, performed a review in which they definitively differentiated verrucous hemangioma from angiokeratoma circumscriptum, with which there had previously been a degree of confusion.

Most authors consider verrucous hemangioma to be a malformation of the dermal capillaries^{2,3} and defend the

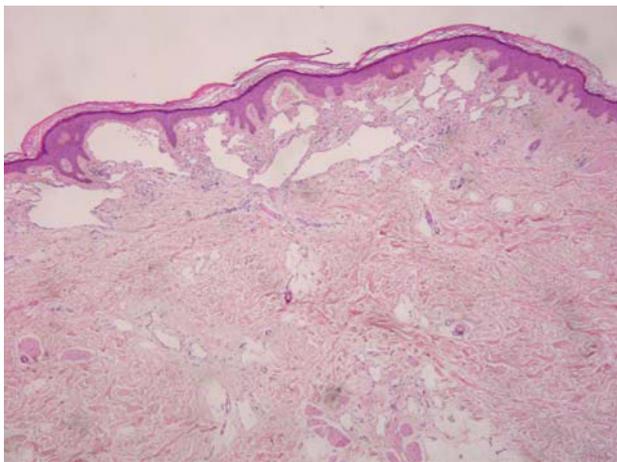


Figure 3. Acanthotic epidermis with hyperkeratosis and papillomatosis; dilated vascular structures are present in the superficial dermis. Hematoxylin-eosin, $\times 40$.

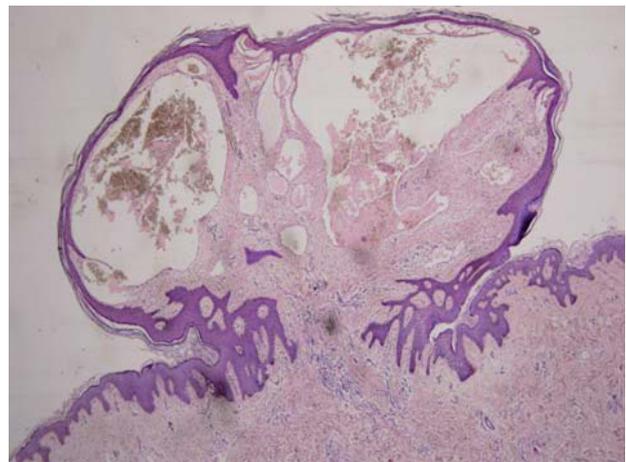


Figure 4. Acanthotic epidermis with hyperkeratosis and papillomatosis; large, dilated vascular structures in the superficial dermis. Hematoxylin-eosin, $\times 10$.

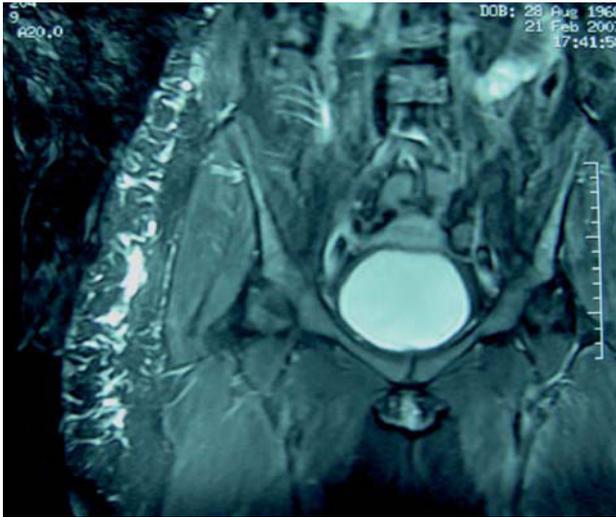


Figure 5. Nuclear magnetic resonance: tortuous, tubular images of reticular morphology, extending across the full thickness of subcutaneous cellular tissue.

use of the term hyperkeratotic vascular malformation^{2,4} to refer to this type of vascular disorder. However, based on histopathologic and immunohistochemical characteristics, other authors are less willing to include verrucous hemangioma within the group of vascular malformations or tumors.⁵

Verrucous hemangioma presents at birth or during infancy, usually on the lower limbs (95%). The lesions tend to be unilateral, well-defined, discrete, grouped, bluish-red, soft, and compressible; they vary between 4 mm and 7 cm in diameter and small satellite lesions are often present.⁶ Complications such as secondary bleeding and infections are common,³ with the result that lesions become hyperkeratotic and acquire a verrucous surface.⁶

Histologically, verrucous hemangioma presents an epidermis with irregular acanthosis and hyperkeratosis.⁵ The abnormal vessels are located in the dermis and hypodermis, and extend along the vertical vascular channels with almost no involvement of the reticular dermis. The vessels are round, with thick walls and a multilamellar basement membrane. The dilated vessels of the papillary dermis often contain blood, are thin-walled, and have a vertical orientation, whereas the deeper vessels may contain blood or be empty. Immunohistochemically, the endothelium shows focal positivity for type 1 glucose transporter and low-level reactivity for mindbomb homolog 1. In contrast, it does not stain with D2-40.

The differential diagnosis includes Cobb syndrome, angioma serpiginosum, lymphangioma circumscriptum, cutaneous keratotic hemangioma, blue rubber bleb nevus, papillomas, and tumors including melanoma.⁶ However,

the main differential diagnosis must be made with angiokeratoma circumscriptum. There is considerable confusion in the literature about when to use these terms, and some authors consider them to be variants of the same disease.^{7,8} It is true that they are clinically similar lesions, but certain details can help to differentiate them: verrucous hemangioma is typically present at birth, whereas angiokeratoma circumscriptum is usually an acquired dermatosis; verrucous hemangioma is usually a solitary lesion that varies from 1 to 7 cm in diameter and is often surrounded by smaller satellite lesions; angiokeratoma circumscriptum, on the other hand, is typically formed of punctate lesions that vary between 1 and 5 mm in diameter and that occasionally coalesce to form plaques several centimeters across.⁹ The main histological difference is that verrucous hemangioma extends into the hypodermis whereas angiokeratoma circumscriptum is limited to the superficial dermis. In fact, some authors state that to classify a lesion as angiokeratoma, there must be no involvement beyond the papillary dermis.¹⁰

Imaging studies such as nuclear magnetic resonance help us to see the deeper tissues in this type of lesion.^{11,12} This is not only of scientific value, but is also important therapeutically as, while angiokeratoma circumscriptum can be treated with the usual physical methods such as electrocoagulation, cryotherapy, and argon laser, verrucous hemangioma requires wide excision to avoid possible recurrence.^{6,9}

In view of this situation, we consider that biopsy of sufficient depth is required in this type of lesion, as a superficial tissue sample may lead to an erroneous initial diagnosis, as occurred in our case, with the consequent inappropriate treatment. Completing the study with imaging techniques such as magnetic resonance enables the superficial and deep features of these angiokeratoma-type lesions to be seen.

Conflicts of Interest

The authors declare no conflicts of interest.

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