Late onset angioma serpiginosum of the breast with co-existing cherry angioma

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ABSTRACT

Angioma serpiginosum is a cutaneous vascular nevoid disorder that presents as red, nonblanchable and grouped macules distributed in a serpiginous pattern and resembles purpura. It usually begins in childhood and commonly affects the lower limbs and the buttocks. Late onset of the lesions is unusual. We report a 46-year-old female who presented with progressively extending lesions on her breast since preceding four years. She also had a cherry angioma overlying the lesions, an observation hitherto not reported so far.

Key words: Angioma serpiginosum, breast, cherry angioma, late onset

INTRODUCTION

Angioma serpiginosum is a benign vascular nevoid disorder with proliferation and ectatic dilatation of capillaries in the papillary dermis. It predominantly affects women with an onset in childhood and is often asymptomatic. Most cases are sporadic but familial cases suggesting an autosomal dominant inheritance have also been described. We report a case of late onset angioma serpiginosum localised to the breast, an unusual site.

CASE REPORT

A 46-year-old female patient presented to the dermatology out-patient clinic with an asymptomatic, progressive red eruption on the right breast of 4 years duration. She did not provide a history of bleeding disorder, preceding trauma, or contact allergy prior to the eruption of the lesions. Her medical and family history was noncontributory. Clinical examination revealed multiple punctate macules in a serpiginous pattern against a bluish background, grouped at places, located on the lateral half of the right breast. There were no similar lesions elsewhere on the body. Diascopy using a glass slide revealed nonblanchable lesions. A 3 mm, red, soft papule was noted overlying these lesions at their lower extent [Figure 1].

The differentials considered were angioma serpiginosum, unilateral nevoid telangiectasia, pigmented purpuric dermatoses and telangiectasia macularis eruptiva perstans.

Epiluminescence microscopy with Heine Delta 20 dermatoscope (Heine Optotechnik, Herrsching, Germany) revealed multiple well demarcated oval to round red lagoons [Figure 2].

Histopathological examination of the nonblanching punctate macules showed a normal to mildly orthohyperkeratotic epidermis with dilated thin walled capillaries in the papillary dermis. There was no evidence of extravasation of erythrocytes, inflammatory cell infiltrate or deposition of hemosiderin in the surrounding tissue [Figure 3]. Periodic acid-Schiff (PAS) stain showed a thick cuff of amorphous acidophilic PAS-positive diastase-resistant material surrounding the dilated vessels [Figure 4]. The red papule overlying the punctate macules revealed a well delineated papillary dermal lesion composed of closely placed ectatic thin walled capillaries engorged with erythrocytes that was consistent with a diagnosis of cherry angioma [Figure 5]. The clinical examination complemented by epiluminescence microscopy and histology confirmed the diagnosis of angioma serpiginosum. Patient was counselled about the benign nature of the disease. She was advised ophthalmic examination, which was refused and no further treatment was sought.

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DISCUSSION

First described by Hutchinson in 1889 and named by Radcliffe-Crocker in 1893, angioma serpiginosum consists of multiple red, minute, nonblanchable and grouped macules, resembling purpura, in a serpiginous or gyrate pattern with a background of erythema or violaceous hue. These progressively extend over months to years.\(^1\) The erythematous or violaceous background hue may be due to dilatation of the subpapillary venous plexus.\(^4\) The eruption usually affects teenage females and in 90% cases has its onset before the age of 16 years.\(^5\) It commonly affects the lower extremities and buttocks and is often asymmetric.\(^6\) Any anatomic site with exceptions of the mucocutaneous junctions, palms and soles can be affected, though there has been a case report describing plantar involvement\(^7\) as well as reports of disseminated distribution.\(^2,4,8-10\)

Majority of cases occur in females and are of childhood onset.\(^2\) In view of female preponderance and progression of lesions in pregnancy, raised levels of estrogens have been postulated in the etiology.\(^6\) The role of hormonal stimuli has been refuted by the absence of estrogen-progesterone receptor stimulation.\(^5\) It has been proposed to represent a nevoid vascular malformation or a vascular neoplasm.\(^2,8\) Though benign and asymptomatic, angioma serpiginosum can be cosmetically disfiguring. No topical medications have proven effective in the treatment. Excellent therapeutic results have been achieved using pulsed dye laser.\(^1\)

Epiluminescence microscopy reveals demarcated red lagoon appearance due to the presence of increased and dilated vascular spaces in the papillary dermis and can help distinguish the condition from purpuric dermatoses on the basis of typical lagoon pattern.\(^11\) Histopathological findings include a normal epidermis, dilated thin walled capillaries in the papillary dermis without an extravasation of red blood cells or deposition of hemosiderin in the surrounding tissue. The capillary walls may be thickened due to deposition of PAS positive material around blood vessels.\(^12\) The absence of dermal mast cell increase, hemosiderin deposition and red blood cell extravasation in our case ruled out the differential diagnoses that were considered.

Cherry angioma is the most common acquired vascular proliferations of the skin. It was first described by Campbell de Morgan in 1872.\(^13\) Generally developing after the third decade, the number and size of these lesions increase with age. There is no sex predilection. They are usually asymptomatic, but may bleed with trauma. They are commonly located on the trunk or proximal extremities. Clinically, early lesions appear as flat, red macules that look like petechiae that later evolve into 1-5 mm red papules. The etiology of cherry angiomas is largely unknown. These are known to be related to chemical exposures, with liver transplants, graft-versus-host disease,
and secondary to cyclosporine treatment. In the absence of any known inciting factors, it is assumed that our patient had an incidental onset of solitary cherry angioma or as a result of chronological ageing.

Histological examination of cherry angiomas shows numerous, newly formed capillaries in a lobular pattern in the papillary dermis. The capillaries have narrow lumina and prominent endothelial cells. They are produced by tortuous dilatation of capillary loops in dermal papillae. Each abnormally dilated loop is connected to the neighboring loop by tortuous vascular channels. Ultra structurally, it is composed of both venous capillaries and postcapillary venules.

Angioma serpiginosum usually begins before puberty. The chest is an unusual site of involvement. There have reports of two female patients with chest wall involvement with the onset at 7 and 9 years of age. Chest involvement has also been described as a part of disseminated lesions in three patients. Two of these patients were beyond 50 years of age at presentation with the onset of their lesions in early childhood. Angioma serpiginosum has been described to be associated with angiokeratoma of the vulva in contiguous areas. However, there has not been any report of cherry angioma in association with lesions of angioma serpiginosum or overlying the lesions.

Our patient presented with a solitary cherry angioma superimposed on the angioma serpiginosum with the onset beyond 40 years of age and on the breast that is an unusual site for the latter. To the best of our knowledge, there has been no reported case of this entity with late onset lesions in the English language literature. Co-existence of cherry angioma is assumed to be a result of chronological ageing process or may be secondary to a common vascular origin of both these lesions.
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