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Cherry Angioma with a Garland of Lichen Sclerosus: The Mystery Continues

Liken Sklerozus ile Çevrili Kiraz Anjiyomu: Gizem Devam Ediyor

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ABSTRACT Cherry angiomas (CA) are common acquired benign vascular skin lesions of unknown etiology that present as small red soft papules mainly over trunk and arms. Lichen sclerosus et atrophicus (LSEA) is a chronic disorder of unknown etiology affecting ano-genital skin in women and the genital skin in men. Coexistence of CA with LSEA has rarely been reported in the literature. Lesions of LSEA completely surrounding a CA like a garland has not been described before. In this article, the author has described a unique case where a CA is completely surrounded by the lesions of LSEA in a garland like fashion. The rarity of this phenomenon obliged the author to report this case.

Keywords: Cherry angioma; extragenital lichen sclerosus; lichen sclerosus; lichen sclerosus et atrophicus

ÖZET Kiraz anjiyomu [cherry angioma (CA)], çoğunlukla gövde ve kollarda küçük kırmızı yumuşak papüller olarak ortaya çıkan, etiyolojisi bilinmeyen, yaygın, edinsel, iyi huylu vasküler deri lezyonlarıdır. Liken sklerozus ve atrofikus [lichen sclerosus et atrophicus (LSEA)], kadınlarda anogenital deriyi ve erkeklerde genital deriyi etkileyen, etiyolojisi bilinmeyen, kronik bir hastalıktır. Literatürde CA ile LSEA birlikteliği nadiren bildirilmiştir. CA'yı bir çelenk gibi tamamen çevreleyen LSEA lezyonları daha önce tanımlanmamıştır. Bu makalede yazar, kiraz anjiyomunun çelenk benzeri bir tarzda tamamen LSEA lezyonları ile çevrili olduğu özgün bir vakayı anlatmıştır. Bu fenomenin nadirliği, yazarı bu vakayı bildirmeye zorunlu kılmıştır.

Anahtar Kelimeler: Kiraz anjiyomu; ekstragenital liken sklerozus; liken sklerozus; liken sklerozus ve atrofikus

Cherry angiomas (CA) are common acquired benign vascular skin lesions of unknown etiology that present as small red soft papules mainly over trunk and arms. They increase in number with age.[1] Lichen sclerosus et atrophicus (LSEA) is a chronic disorder of unknown etiology affecting ano-genital skin in women and the genital skin in men. Extragenital lesions are seen in about 10% of female patients while isolated extragenital involvement is seen in only 2.5% cases.[2,3] Coexistence of CA with LSEA has rarely been reported.[3] Lesions of LSEA completely surrounding a CA like a garland has not been reported till date.

A 40-year-old female presented with multiple asymptomatic whitish lesions on her back which she noticed 6 months ago. She denied any past or family history of the same. On examination, there were multiple small rounded hypopigmented/depigmented atrophic macules and papules of variable size (2-6 mm) which were discrete at some places while coalescing with each other to form plaques at other (Figure 1a). At the center of her back, a dome shaped, cherry red colored polypoid papule 0.8-1 cm in size was completely surrounded by these white colored atrophic macules (Figure 1b). On questioning, she mentioned that this red papule has been there for few years without causing any concern for her. Rest of the dermatological examination including hair, nails and mucosae (oral/genital) was unremarkable. Systemic examination was non-contributory. Examination with Wood's lamp did not show accentuation or fluorescence ruling out the

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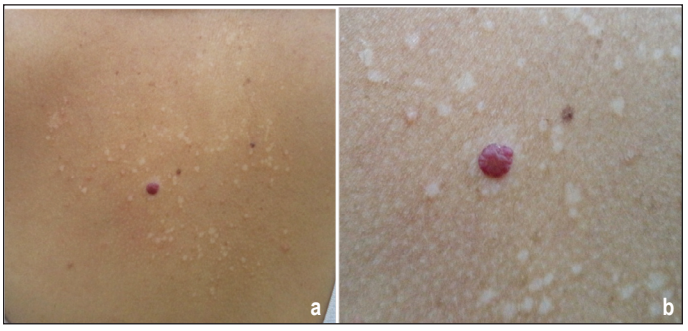

FIGURE 1: Cherry angioma surrounded by multiple small rounded hypopigmented/depigmented atrophic macules and papules of variable size (a) close view of central cherry angioma completely surrounded by whitish atrophic lesions of lichen sclerosus et atrophicus on all the four quadrants (b).

possibility of vitiligo or pityriasis versicolor. Since the patient had no concern for the reddish papule, a 4 mm punch skin biopsy was taken from the depigmented lesion, which showed atrophic epidermis with orthohyperkeratosis, pronounced edema and hyalinized collagen bundles in papillary dermis and moderate lymphoplasmocytic infiltrate in mid dermis (Figure 2). Based on history, clinical examination and further supported by histopathological findings, a diagnosis of CA with LSEA was made.

CA with a halo needs to be differentiated from the present case. Karaman described pale halo formation in 2% of CA's examined by him. However, such halo consists of sharply defined depigmented rings and is macular in nature.[4,5] In the present case, there was change in skin texture and the whitish garland was irregular with variable widths in different quadrants.

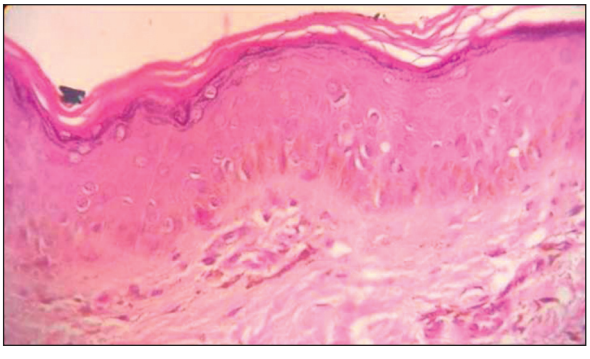

FIGURE 2: There is mild to moderate orthohyperkeratosis, atrophic epidermis, follicular plugging with a clear zone at epidermo-dermal junction. Dermis shows mild to moderate inflammatory infiltrate with hyalinized and dense collagen bundles (H&E, x40).

Despite meticulous literature review, the author could find only single report where CA coexisted with extragenital LSEA. Ingram et al. reported two female patients with extragenital LSEA affecting their axillae and lower abdomen. On examination, both were found to have several hundred CA in a segmental distribution involving left thigh and flank of first patient, and right abdomen and back of the second.[3] However, in their case LSEA and CA were present in different anatomical regions but in this case LSEA completely surrounded the CA like a garland, a phenomenon not reported before.

A probable reason for this close proximity between CA and extragenital LSEA may reflect a shared defect in the cutaneous microcirculation. Extragenital LSEA has been reported in association with limited systemic sclerosis and morphea, both of which have recognized defect in microvasculature.[6-8] Argument against this link is that macular telangiectasia seen in systemic sclerosis occur due to the dilatation of post-capillary venules while in CA there is dilatation of capillary loops.[1] Another possibility is that both CA and extragenital LSEA have been reported in segmental distribution pointing to genetic mosaicism.[3,9] Thus, an attempt to link the two entities etiologically remains elusive. Nevertheless, such observations are compelling us to contemplate about possible relationship between the two and hence mystery continues.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

This study is entirely author's own work and no other author contribution.

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