

Cutaneous Leiomyoma Mimicking a Keloid

Dear Editor,

Cutaneous leiomyomas (CL) are rare, benign smooth muscle tumors of the skin (1). There are 3 subtypes with different origins and histopathologic features: piloleiomyoma, genital leiomyoma, and angioleiomyoma (2). Pilar leiomyoma is the most common subtype originating from arrector pili muscles of pilosebaceous unit. It presents as painful solitary or multiple papulonodules (2,3).

A 30-year-old woman presented to our outpatient clinic with numerous painless, itchy papules on her gluteal region that had been present for 10 years. Dermatologic examination revealed red-brown, smooth, grouped papulonodules on bilateral gluteal regions (Figure 1). These lesions had appeared after intramuscular injections and had increased in number. Family history was unremarkable. A punch biopsy was performed with pre-diagnoses of keloid and tumoral infiltration. Histopathologic examination showed neoplastic infiltration with large bundles of spindle-like smooth muscle cells with acidophilic cytoplasm under epidermis (Figure 2). Neoplastic cells were stained by smooth muscle markers actin and caldesmon (Figure 3). Based on the clinical and histopathological findings the diagnosis of pilar leiomyoma was established. Pelvic and renal ultrasonographic examinations were normal. The patient's lesions were asymptomatic except for mild itching and she is currently in follow-up without any treatment.

Pilar leiomyomas mostly manifest around the ages of 10 to 30 and are located on the trunk and ex-

tensor surfaces of the extremities. Lesions are firm, red-brown or skin-colored papulonodules with diameters varying from several mm to 2 cm (2). Differential diagnosis includes dermatofibroma, neurofibroma, smooth muscle hamartoma, neuroma, adnexal tumors, and painful papulonodular lesions such as glomus tumor (1,2).

Our case clinically resembled keloid with red-brown, stiff nodules with epidermal thinning. In the literature, a patient with cutaneous pilar leiomyoma was diagnosed with eruptive keloid and treated with cryotherapy and intralesional steroid injections before histopathologic verification of pilar leiomyoma. He had multiple painless, red-purple papulonodules on the chest and arms (3). The case of a 53-year-old man with a history of multiple firm and painful lesions on the back showing segmental distribution and diagnosed with keloid-like leiomyoma was also reported (4). CL should be considered in the differential diagnosis of keloid-like lesions with atypical location and that are resistant to treatment.

Cutaneous leiomyomas have different clinical presentations and many differential diagnoses, but CL can be diagnosed by histopathological examination. In all CLs, histopathologic examination shows bundles of spindle-shaped smooth muscle cells with eosinophilic cytoplasm, a cigarette-like nucleus, and a perinuclear halo. Smooth muscle markers actin and desmin are routinely positive. Histopathologic examination in our case also revealed bundles of



Figure 1. Red-brown, smooth, grouped papulonodules on the upper outer quadrant of the bilateral gluteal regions.

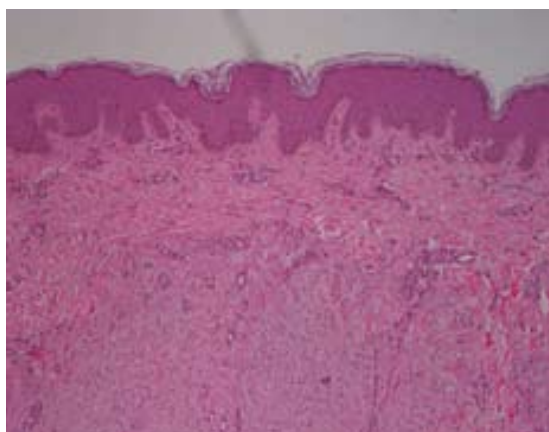


Figure 2. Mesenchymal tumor with bundles of spindle-like cells under the epidermis (hematoxylin and eosin $\times 100$).

spindle-like smooth muscle cells with large acidophilic cytoplasm; smooth muscle markers actin and caldesmon were positive.

While solitary lesions are frequently sporadic cases, multiple lesions may be related to hereditary conditions such as Reed's syndrome (multiple cutaneous and uterine leiomyomatosis), hereditary leiomyomatosis, and renal cell cancer (2). These two hereditary conditions have been reported to be associated with a heterozygous germline mutation in fumarate hydratase gene (4). Our patient was considered a sporadic case due to lack of family history and uterine leiomyoma and normal renal ultrasonography.

Treatment of CL depends on the number of lesions and presence of symptoms (1). Surgical excision is the gold standard in the treatment of solitary and self-limiting lesions (2). However, recurrence can be more commonly observed in patients with multiple lesions (1). Drugs targeting smooth muscle contraction such as nifedipine, nitroglycerin, and phenoxybenzamine are recommended for pain management. Methods such as cryotherapy and carbon dioxide laser ablation have been tested but their efficacy was found to be limited (1,2). In our patient, lesions were asymptomatic and few in number; we thus suggested follow-up without any treatment.

CL are rare benign smooth muscle tumors of the skin. They are difficult to diagnose by clinical evaluation, but the diagnosis can be established by histopathologic examination. In patients with atypical keloid-like papulonodular lesions like our patient, pilar leiomyoma should be considered and histopathologic examination should be performed for the diagnosis.

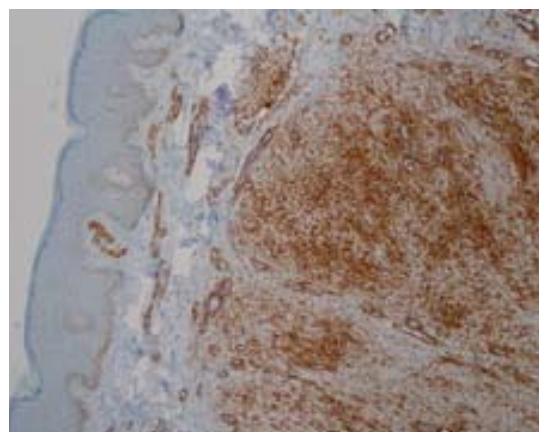


Figure 3. Cytoplasmic positivity of caldesmon in spindle-like cells (E89 $\times 100$).

References:

1. Holst VA, Junkins-Hopkins JM, Elenitsas R. Cutaneous smooth muscle neoplasms clinical features, histologic findings, and treatment options. *J Am Acad Dermatol.* 2002;46:477-90.
2. Malik K, Patel P, Chen J, Khachemoune A. Leiomyoma cutis: a focused review on presentation, management, and association with malignancy. *Am J Clin Dermatol.* 2015;16:35-46.
3. Morariu SH, Suci M, Badea MA, Vartolomei MD, Buicu CF, Cotoi OS. Multiple asymptomatic cutaneous pilar leiomyoma versus spontaneous eruptive keloids - a case report. *Rom J Morphol Embryol.* 2016;57:283-7.
4. Chia MW, Tay YK, Tan SH. Multiple leiomyoma as a possible sign of aggressive renal cancer. *Int J Dermatol.* 2008;47:1157-8.

Emine Boyuk¹, Zeynep N. Saracođlu¹, Deniz Arık²

¹Department of Dermatology, Eskişehir Osmangazi University, Faculty of Medicine, Eskişehir, Turkey,

²Department of Pathology, Eskişehir Osmangazi University, Faculty of Medicine, Eskişehir, Turkey

Corresponding author:

Emine Boyuk, MD

Department of Dermatology,
Eskişehir Osmangazi University,
Faculty of Medicine, Eskişehir,
Turkey

emineboyuk@gmail.com

Received: February 15, 2018

Accepted: May 15, 2020