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# Painful erythematous, infiltrated papulonodules in a middle aged man

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## Abstract

Cutaneous leiomyomas (CL) often present as painful tumors in the skin. Herein we report an adult man who presented with multiple, erythematous papulonodules in both segmental and non-segmental distribution.

Keywords: painful skin tumor, leiomyoma cutis

## Case Synopsis

A 48-year-old man presented with multiple papulonodules over the back and face for 25 years. He initially noticed grouped lesions on the right flank; subsequently similar lesions appeared over the left flank and right side of the face. It was asymptomatic initially, but gradually, 2 years prior to presentation, he started experiencing a burning sensation, which had worsened in the last 15 days. He did not have any aggravating or relieving factors. Other significant medical illness included diabetes and hypertension for which he was taking medications. Cutaneous examination revealed multiple, grouped, erythematous, infiltrated papules and nodules over the right preauricular, right submandibular, right infrascapular and lumbar, and left infrascapular regions. A few discrete papules were noted over the abdomen (**Figure 1**). Lesions over the left infrascapular region were arranged in a linear pattern (**Figure 2**). Examination of peripheral nerves were normal. Slit skin smear (SSS) from the standard sites and lesions were negative. Routine hematological and biochemistry investigations showed raised sugar and cholesterol levels. Abdominal ultrasonography was



**Figure 1.** Multiple, grouped, erythematous, infiltrated papules and nodules over the right infrascapular and lumbar region.

normal. An excisional biopsy of the representative lesion was taken and sent for histopathological examination.

## Histopathological findings

Histopathology examination showed a poorly circumscribed, non-capsulated nodule in the dermis composed of interlacing fascicles of spindle cells with blunt ended elongated nuclei and eosinophilic cytoplasm (**Figure 3**). A Masson trichrome stain revealed bright red cytoplasmic staining of tumor cells. Immunohistochemical studies were performed and cells stained positive for smooth muscle actin (SMA) and negative for S100. A final diagnosis of cutaneous leiomyoma was made.

## Case Discussion

Cutaneous leiomyomas (CL) are benign soft tissue



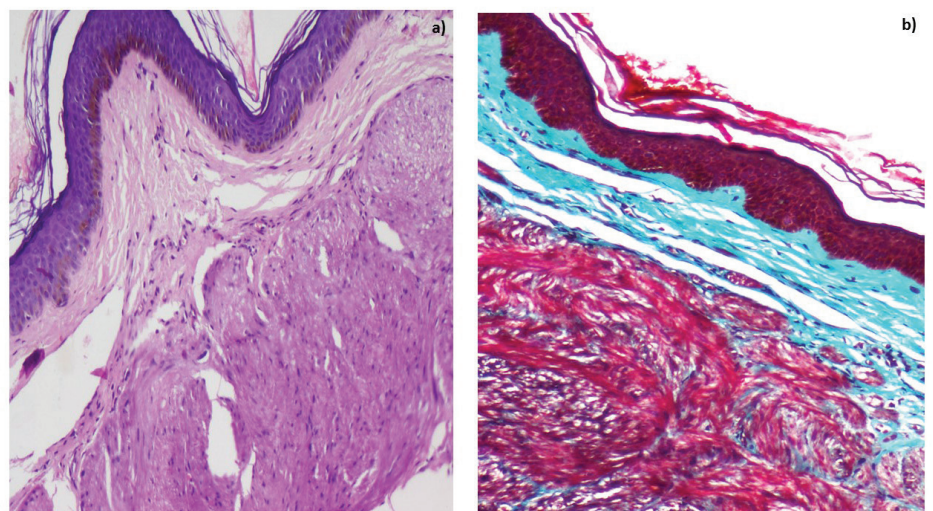
**Figure 2.** a) Infiltrated papulonodular lesions in a dermatomal pattern over the left infrascapular region and, b) over the preauricular region.

neoplasms that arise from the smooth muscle bundles. Skin is the second most common location for leiomyoma after uterus. CL account for 75% of extrauterine leiomyomas [1]. According to their site of origin, leiomyomas may be classified into piloleiomyomas, angioleiomyomas, and genital leiomyomas [2]. Piloleiomyomas are derived from the arrector pili muscle of the hair follicles, whereas angioleiomyomas are derived from the vascular smooth muscle. Genital leiomyomas originate from the dartoic, vulvar, or areolar smooth muscles [3]. CL usually occur in patients in the age group of 10-30 years with equal frequency in both genders [4]. It usually present as small red-brown, firm papules on the trunk or extremities. Lesions may be solitary or multiple; solitary lesions usually occur on the extremities, whereas multiple lesions are seen more commonly over the trunk (known as leiomyomatosis). Multiple lesions may occur in a disseminated fashion or rarely may be segmental (zosteriform leiomyoma) [5]. Sometimes affected patients may have multiple lesions arranged in groups in a regional distribution; more than one body site may be affected [6]. Our patient had multiple, grouped lesions involving more than one body region as well as lesions arranged in a segmental distribution.

Patients often experience pain that may be spontaneous or may be aggravated by cold, pressure, or emotion [7]. Although the pathophysiology of pain is still unknown, it may result from local pressure on cutaneous nerves by the tumor. Other proposed hypotheses for pain include infiltration of mast cells and local vasoconstriction leading to muscle contraction. Segmental and multiple disseminated lesions tend to cause more pain [5, 8].

Histologically, leiomyomas are composed of well differentiated interlacing bundles of smooth muscle fibers with elongated nuclei giving a cigar-shaped appearance. Varying amounts of collagen bundles are intermingled with smooth muscle bundles. Smooth muscles stain dark red with the Masson trichrome stain [6, 7]. On immunohistochemical staining, the muscle spindle stains positive for smooth muscle actin [5].

CL should be differentiated from neurofibroma, eccrine spiradenoma, dermatofibroma, and angiolipoma. It may be associated with myomas in other areas such as the gastrointestinal tract, retroperitoneum, or uterus in females (Reed syndrome). An association of CL with renal papillary cell cancer and uterine leiomyoma has been described [9]. This is inherited in an autosomal dominant fashion and is owing to



**Figure 3.** a) Photomicrograph showing well circumscribed dermal tumor composed of intersecting fascicles of plump spindle shaped cells (H&E, 200x), b) Section shows bright red cytoplasmic staining in tumor cells (Masson's trichrome, 200x).

mutation in fumarate hydratase, an enzyme that catalyzes the conversion of fumarate to malate in the Krebs cycle [10]. Therefore, a detailed history, examination, and investigations should be carried out in patients with multiple piloleiomyomas [8]. Our patient denied any history of systemic symptoms; routine urine and biochemical investigations were normal. Abdominal ultrasound was normal, thus ruling out the co-existence of Reed syndrome in our patient.

Surgical excision is the treatment of choice for solitary CL. However it is not practical for patients with extensive or multiple lesions, in whom medical management has been attempted. Treatment with various pharmacological agents prescribed include nifedipine, alpha adrenoreceptor blockers (phenoxybenzamine), nitrates, analgesics, antidepressants, and gabapentin. Physical modalities like CO2 laser ablation, liquid nitrogen cryotherapy, and electrocoagulation have been attempted in a few cases with success [11]. Recently, botulinum toxin injection has been used with promising results [12].

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