CASE REPORT

Zosteriform Cutaneous Leiomyoma: A Rare Cutaneous Neoplasm

Arfan-ul-Bari

ABSTRACT

Cutaneous leiomyomas are firm, round to oval, skin-coloured to brownish papules and nodules that may present as a solitary, few discrete or multiple clustered lesions. Different uncommon patterns of multiple leiomyoma distribution have been noted as bilateral, symmetrical, linear, zosteriform, or dermatomal-like arrangement. One such rare presentation was seen in a 23-year-old patient who presented with zosteriform skin coloured, occasionally painful cutaneous lesions over left shoulder region. Histopathology confirmed the diagnosis of cutaneous leiomyoma. He was symptomatically managed with non-steroidal anti-inflammatory agents and topical capcicum cream. Case is reported here due to rare occurrence of this benign cutaneous neoplasm in an atypical pattern and on uncommon site.

Key Words: *Cutaneous leiomyoma. Zosteriform cutaneous leiomyoma. Multiple cutaneous leiomyomas.*

INTRODUCTION

Cutaneous leiomyomas typically present as reddish-brown or skin-coloured, firm, often painful nodules, preferentially over the face, neck, trunk, or extremities. Lesions on the trunk can be multiple, disseminated, zosteriform, or segmental in distribution while lesions over the extremities tend to be solitary. Patients usually complain of hypersensitivity to light touch and cold temperature which may be spontaneous or induced by trauma, and pressure. Some may report burning, pinching, or stabbing pain in lesions. Segmental and multiple disseminated lesions tend to cause more pain.[1-3] Clinically, leiomyoma lesions may look similar to other benign cutaneous neoplasms and diagnosis can only be confirmed by characteristic histology of the lesion. Histopathology reveals interlacing bundles of smooth muscle, collagen fibers and variable amount of lymphocytic and mast cell infiltrate occupying whole of reticular dermis.[1,4]

The treatment of leiomyomas is not satisfactory. Surgical excision is the therapy of choice in solitary, painful lesions but chances of recurrence in case of multiple, painful lesions make surgery less practical. Various systemic therapies have also been tried with variable results, such as calcium channel blockers, nitrates, analgesics, antidepressants, gabapantine. Liquid-nitrogen cryotherapy and CO_2-laser ablation have also been used with good results.[1,5-7]

The object of presenting this case is to highlight the occurrence of this interesting benign tumour of skin in an atypical manner and on uncommon site which was adequately managed with conservative treatment.

Department of Dermatology, Combined Military Hospital, Bahawalpur.

Correspondence: Lt. Col. Dr. Arfan-ul-Bari, Consultant Dermatologist, Combined Military Hospital, Bahawalpur. E-mail: albariul@gmail.com

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CASE REPORT

A 23-year-old man presented with multiple erythematous papulo-nodular lesions of 15-year duration on the left half of upper chest. He had developed few similar but isolated lesions on left shoulder and left upper arm over the past 2 years. There were frequent brief episodes of pain in the lesions, aggravated on exposure to cold, touch, and physical stress. His general physical and systemic examinations were within normal limits.

Dermatological examination revealed multiple erythematous, indurated plaques measuring approximately 13 x 7 cm and composed of papules and nodules of different sizes. Lesions were arranged in zosteriform pattern over left sub-clavicular region along dermatomal distribution of T-2 segment with clear midline demarcation. Few solitary satellite lesions were present on the left shoulder and left upper arm (Figure 1a).

An excision biopsy of one of the nodules was done. Histopathology revealed a tumour composed of irregularly arranged bundles of smooth-muscle fibers, with elongated cigar-shaped nuclei and rounded ends, interlaced with variable amounts of collagen infiltrating the full thickness of the dermis (Figure 1b). Von Gieson stain was also positive for smooth muscle.

He was symptomatically managed with non-steroidal anti-inflammatory drug (diclofenac sodium) twice daily

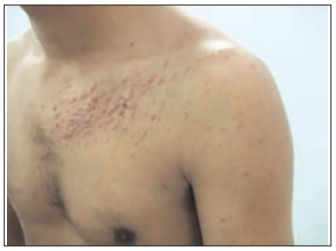

Figure 1(a): Zosterifom distribution of multiple cutaneous leiomyoma lesions along T-2 segment on left upper chest.

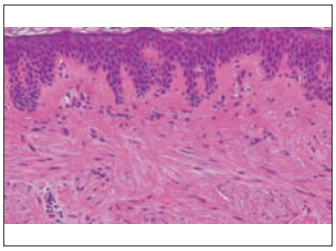

Figure 1(b): Bundles of smooth muscle fibers with elongated cigar shaped nuclei interlaced with bundles of collagen in the dermis (Hematoxylin-Eosin, x 200).

after meals and topical capcicum cream twice daily application. After 10 days, diclofenac sodium was stopped and was advised to apply capcicum whenever, he experienced pain or burning sensation. For the last 4 months, he is asymptomatic.

DISCUSSION

Leiomyomas are benign tumours of the smooth muscle arising from the arrector pili muscle, the muscular layer of the blood vessels or the tunica dartos and accordingly named as piloleiomyoma, angioleiomyoma and dartoic leiomyoma. Piloleiomyoma lesions appear as isolated elements, in groups, or with a metameric distribution. They can occur in different locations such as the extensor surfaces of the limbs, the trunk, or the face. The most common clinical presentation is in the form of solitary lesions. The lesions often cause pain in response to cold or pressure. Older lesions may increase in size and new lesions develop over time and growth is usually gradual.[1,2] Different patterns of multiple leiomyoma distribution have been noted as bilateral, symmetrical, clustered and linear. Zosteriform, segmental or dermatomal-like arrangement of lesions is a rare occurrence.[2,3] The lesions are often sensitive to touch, cold, emotional stress, or spontaneous pain. This pain or hypersensitivity is possibly due to local pressure exerted by the tumour itself on cutaneous nerves or by the infiltrating mast cells in the tumour mass. Muscle contraction is mediated by alpha-adrenergic receptors, which are found in the arrector pili muscle which may also play a role in the induction of pain.[1-4]

The pathogenesis of segmental arrangement of cutaneous leiomyomas is not very much clear. However, it is suggested that as in segmental neurofibromatosis, heterozygosity of a postzygotic mutation is the basis of zosteriform/segmental distribution of skin lesions in case of zosteriform cutaneous leiomyoma.[8] Diagnosing cutaneous leiomyomas in clinical practice may be difficult as macroscopically and symptomatically the lesions closely mimic neurofibroma, eccrine spiradenoma, dermatofibroma, and angiolipoma but diagnosis can be accomplished by microscopic examination of a hematoxylin-eosin stained biopsy of the lesion. Histologically, leiomyomas are generally composed of well-differentiated interlacing bundles of smooth muscle fibers with elongated nuclei displaying blunt ends, appearing as cigar shaped.[1,4]

Leiomyomas may also be associated with Reed's syndrome, polycythemia, visceral involvement (gastrointestinal tract and retroperitoneal area) and renal cell carcinoma.[1,9,10] Therefore, a detailed history, examination, and investigations should be carried out in patients with multiple leiomyomas.

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