CASE REPORT

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Multiple Cutaneous Nodules in Segmental Distribution-Zosteriform Leiomyoma Revisited

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ABSTRACT

Cutaneous leiomyoma are benign smooth muscle tumors of the skin. They present as solitary or multiple flesh colored, occasionally painful skin-colored to brownish papules and nodules. The uncommon pattern of multiple leiomyoma includes linear, zosteriform, or dermatomal like arrangement of lesion. Segmental or zosteriform leiomyoma can occur either along single dermatome (type 1) or with scattered non segmental lesions elsewhere (type 2). Type 2 segmental leiomyomas have been rarely reported in literature and after extensive search in PubMed-Medline database we could find less than ten cases of this variety reported till date from eastern part of India. Here we are reporting a case of type 2 zosteriform leiomyoma for its rarity and its potential association with renal cell carcinoma which required regular follow up.

Keywords: Zosteriform, Leiomyoma, Segmental

Introduction

Cutaneous leiomyomas are benign smooth muscle tumors of the skin which is characterized by painful nodule, that can present either singly or multiply [1]. When multiple they can be arranged in diffuse (disseminated), or segmental (blaschkoid or zosteriform) patterns. Segmental or zosteriform leiomyomas usually occur along single dermatome unilaterally (type 1); or rarely they may be associated with scattered, isolated lesions elsewhere (type 2) [2].

Here we present a case of type 2 zosteriform leiomyoma; as this variety is very rare in occurrence.

Case Report

A 45 year old male patient presented to us with multiple firm painful swelling, mainly on left side of his lower back for the last 20 years. The lesions progressively increased in size over time. He also gives history of pain which was aggravated on exposure to cold. There was no history of any urinary disturbance. Family history was negative. On cutaneous examination skin colored to brown, firm, tender papules and nodules of size 0.5-1 cm present along the posterior aspect of lower back on the left side in segmental or zosteriform pattern. Few scattered lesions were present on upper back, upper and mid portion of his chest and left shoulder (Figure 1). Hair, nail, other mucosae were normal. Systemic examination shows no abnormality. Routine blood examinations were normal. Ultrasonography of lower abdomen was normal and histopathological examination with haematoxylin and eosin stain revealed poorly demarcated interlacing bundles of smooth muscle fibres intermingled with varying amount of collagen (Figure 2, 3, 4). Special stain with Masson's trichome stained the smooth muscle red (Figure 5).

Based on clinical and histopathological findings we diagnosed this case as type 2 zosteriform leiomyoma. Genetic study couldn't be done due to unavailability and financial constraints.

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The patient was counseled about his disease and the therapeutic options were discussed. He refused to undergo any active intervention for his condition. He was given nifedipine (10 mg three times a day) and was kept under regular follow up.

Discussion

Cutaneous leiomyomas comprises approximately 5% of all leiomyomas [3]. According to the site of origin, they are of three types; piloleiomyoma (most common variant derived from arrector pili muscle of hair follicles), angioleiomyoma (derived from vascular smooth muscle) and dartoic leiomyoma (arising from genital smooth muscle) [4]. Cutaneous leiomyomas are more common in adults than in children [3].

Multiple cutaneous leiomyoma is the most common clinical variety, with lesion mainly situated over the trunk and extremities. However rarely they may occur on the tongue or any other part of mouth [4]. The lesions present as small, red brown, firm papule, ranges from few millimeter to 1 cm, and usually fixed to skin but movable over underlying deeper structures [4]. Multiple piloleiomyomas may be inherited in an autosomal-dominant fashion and may be associated

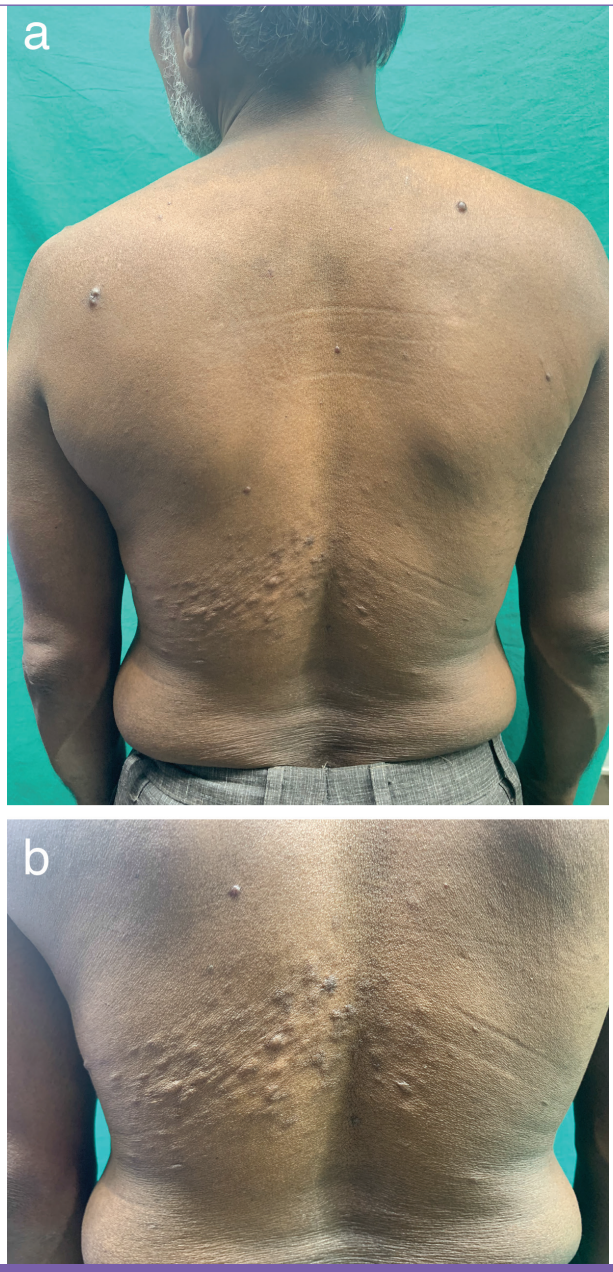

Figure 1. a, b) Multiple cutaneous nodules in zosteriform distribution

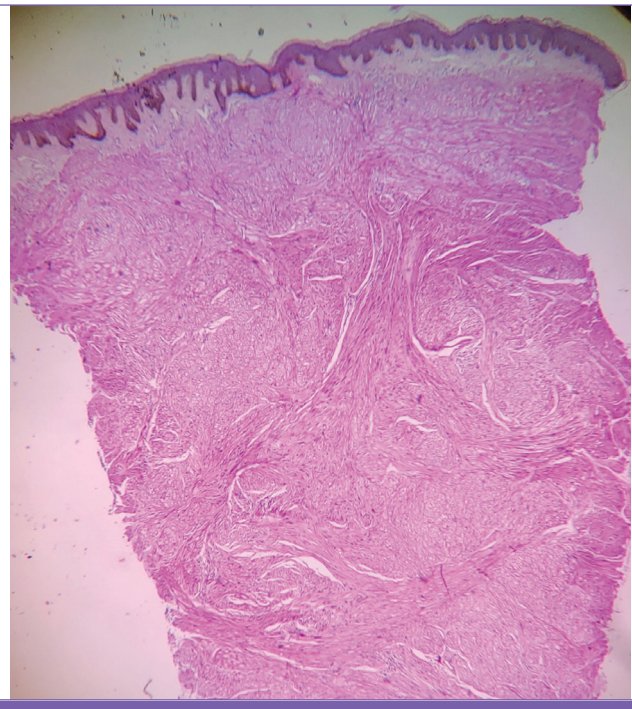

Figure 2. Histopathological examination with haematoxylin and eosin stain under 4x

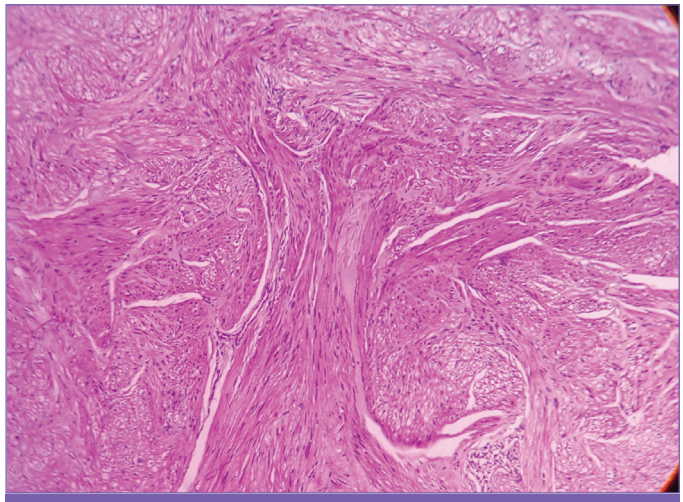

Figure 3. Histopathological examination with haematoxylin and eosin stain under 10x

with uterine leiomyomas and aggressive renal carcinoma, also known as multiple cutaneous and uterine leiomyomatosis or Reed's syndrome and hereditary leiomyomatosis and renal cell carcinoma, respectively [1]. Patients with piloleiomyoma often have pain that may be spontaneous or secondary to cold, pressure, or emotion. The exact mechanism is unknown but the possible explanation could be due to pressure on nerve fibres and abnormal muscle contraction [3]. Our patient also complains of pain on exposure to cold.

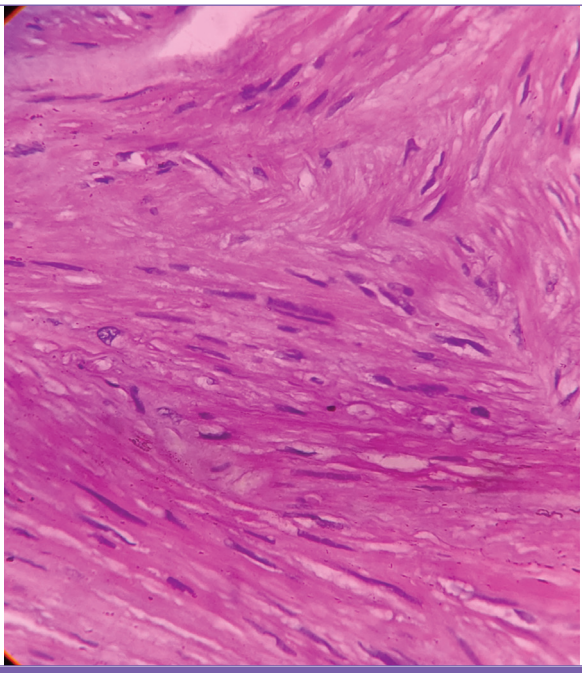

Figure 4. Histopathological examination with haematoxylin and eosin stain under 40x

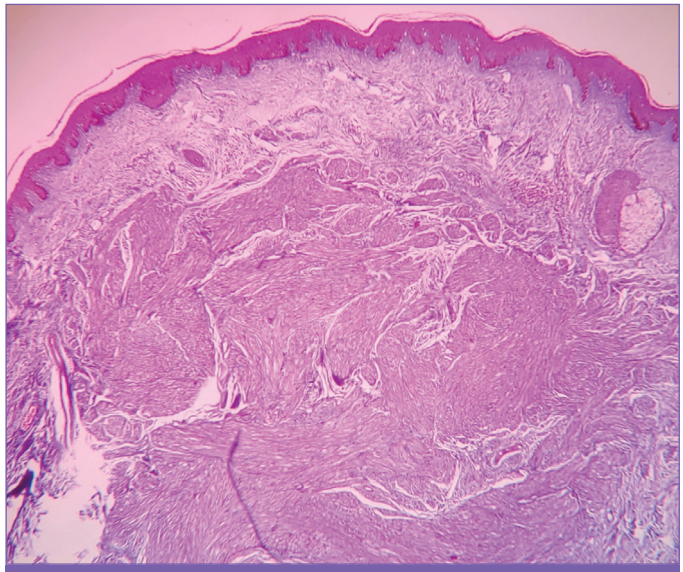

Figure 5. Special stain with Masson's trichrome

Histology of piloleiomyoma is characterized by bundles of smooth muscle arranged in interlacing or whorled pattern with abundant eosinophilic cytoplasm and thin, elongated blunt edged nuclei [5].

Treatment of cutaneous leiomyoma depends on the number of lesions and the presence or absence of symptoms. Different modalities of treatment include medical management with nifedipine, doxazosin, gabapentine, phenoxybenzamine and other alpha-1 blockers; surgical excision if number of lesions few; cryotherapy with liquid nitrogen, CO_2 laser ablation with varying success. The condition may however recur [2,5].

Type 2 segmental leiomyomas have been rarely reported in literature and after extensive search in PubMed-Medline database we could find less than ten cases of this variety reported till date from eastern part of India.

Bandyopadhyay et al. [5] reported a case of 32-year-old man who presented with leiomyoma distributed segmentally over C6 to T8 dermatomes on left side and T11 to L1 dermatomes on right side.

Kudligi et al. [4] reported a case of unilateral multi-segmental leiomyomas along 5th cervical, 6th dorsal, and 1st sacral segments of right side in a 30 year old female.

In conclusion, we are reporting a case of type 2 zosteriform leiomyoma for its rarity and its potential association with RCC which required regular follow up.

Ethics

Informed Consent: Consent form was filled out by a participant.

Peer-review: Internally peer-reviewed.

Authorship Contributions

Concept: S.S., Design: O.R., Data Collection or Processing: S.S., S.D., Analysis or Interpretation: S.Se., Literature Search: S.S., A.H., Writing: S.S., S.D.

Conflict of Interest: No conflict of interest was declared by the authors.

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