Case Report

Solitary cutaneous pilar leiomyoma: A rare entity with review of literature

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ABSTRACT

Cutaneous leiomyomas are uncommon, benign smooth muscle neoplasm and comprise approximately 5% of all leiomyomas. Pilar leiomyoma is the most common type of cutaneous leiomyoma and arises from arrector pili muscle in the skin. Cutaneous pilar leiomyomas present in second and third decades of life as multiple, painful red-brown papulonodular lesions on the trunk or extremities. Clinically this lesion is easily misdiagnosed for other cutaneous conditions. Hence, this lesion should always be taken into consideration in the clinical differential diagnosis of papulonodular skin lesions. Herein, we report a case of 34-year-old male who presented with a small nodule on his left arm. Clinically differential diagnosis was fibroma or neuroma. Surgical excision was performed. Histopathological examination, special stain (Masson's trichrome) and Immunohistochemistry confirmed the diagnosis of cutaneous pilar leiomyoma.

Key words: Arrector pili, cutaneous, pilar

INTRODUCTION

Cutaneous leiomyomas are an uncommon variant of the typical smooth muscle leiomyoma of the uterus, representing only 3–5% of leiomyomas.[1] Skin is the second most common location for leiomyoma after uterus, which accounts for 95% of cases. Cutaneous leiomyomas account for 75% of extra-uterine leiomyomas.[2] Piloleiomyoma is the most common type of cutaneous leiomyoma.[3] Pilar leiomyoma is a rare, benign smooth muscle tumor arising from the arrectorpilimuscle associated with the hair follicles of the skin.[4] They usually appear as red or brown papules <15 mm in diameter. Tumors can be painful due to compression of cutaneous nerves or contraction of fibers within the tumor in case of cold weather or emotional stress.[5] Clinically cutaneous pilar leiomyomas present as solitary or multiple lesions on the trunk or extremities so they may mimic other cutaneous lesions and present a diagnostic challenge. Hence, histopathological examination is the gold standard for accurate diagnosis.

Herein, we present a case of cutaneous pilar leiomyoma in a 34-year-old male, clinically diagnosed as fibroma or neuroma and discuss the review of the literature. Very few cases are reported in the literature.

CASE REPORT

A 34-year-old male presented with 2–3 years history of single nodule on his left arm. He did not give any history of pruritus, burning sensation and tenderness. There was no history of any trauma. His personal and medical history was unremarkable. Family history was also unremarkable for any neoplasm including kidney cancer and uterine fibroids. On local examination, there was single, 2–2.5 cm, a red-brown colored nodule on the left arm. On palpation nodule was soft to firm and nontender. Clinical diagnosis was fibroma or neuroma. Surgical excision was done and sent for histopathological examination. On gross

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examination, we received a single, firm nodule measuring 2 cm. Cut section showed grayish-white whorled area.

Histologically, multiple serial sections showed epidermis with underlying tumor tissue comprising of the proliferation of spindle cells arranged in interlacing and whorled pattern in the deep dermis [Figure 1]. The spindle tumor cells showed blunt-ended cigar shaped nuclei with abundant eosinophilic cytoplasm without cytological atypia or mitotic activity [Figure 2]. Sebaceous gland and hair follicles were also seen. On histopathological examination, pilar leiomyoma was diagnosed. The special stain Masson's trichrome (MTS) stain confirmed the positivity for smooth muscle fibers [Figure 3]. Immunohistochemistry study revealed positivity of smooth muscle actin and negativity of S100 [Figure 4]. On the basis of histopathological examination, special stain MTS and immunohistochemistry final diagnosis of cutaneous pillar leiomyoma was given. No recurrence was observed in the 6 months follow-up of the patient.

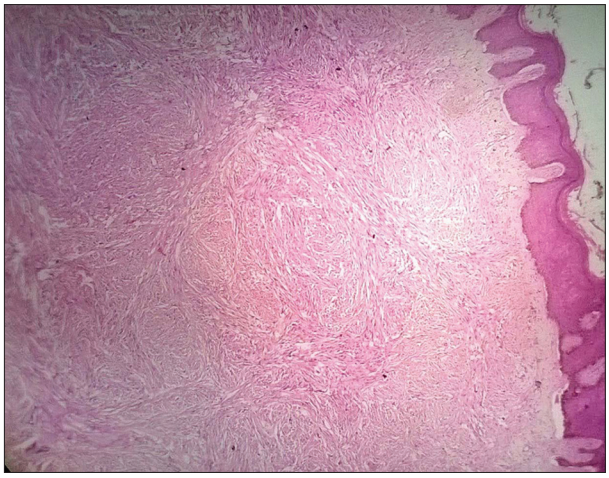

Figure 1: Multiple serial sections from nodule showed epidermis with underneath tumor tissue comprised of proliferation of spindle cells arranged in interlacing and whorled pattern in the deeper dermis. (H and E, ×100)

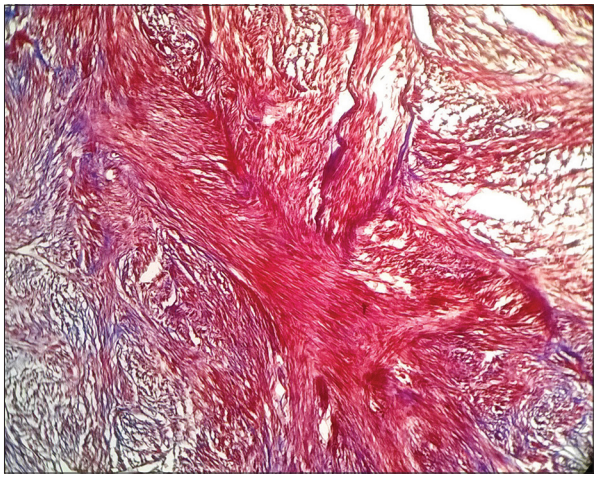

Figure 3: Masson'strichrome stain confirmed the positivity for smooth muscle fibers

DISCUSSION

Cutaneous leiomyomas are benign tumors of smooth muscle bundles. They are divided into five subtypes based on the origin of the smooth muscle: Multiple piloleiomyomas, solitary piloleiomyomas, solitary genital leiomyomas, solitary angioleiomyomas, and leiomyomas with mesenchymal elements.[6]

Multiple piloleiomyomas are the most common type of leiomyomas and are small, firm, red, or brown intradermal nodules affecting the trunk or extremities, but may occur on the face and neck and rarely in the mouth.[6] Multiple cutaneous leiomyomas may be associated with uterine leiomyomas, also known as multiple cutaneous and uterine leiomyomatosis (MCUL), familial leiomyomatosis cutis et uteri, Reed syndrome, or multiple leiomyomatosis.[7] It may also be associated with renal cell carcinoma. The mode of transmission of MCUL is autosomal dominant with incomplete penetrance.[8]

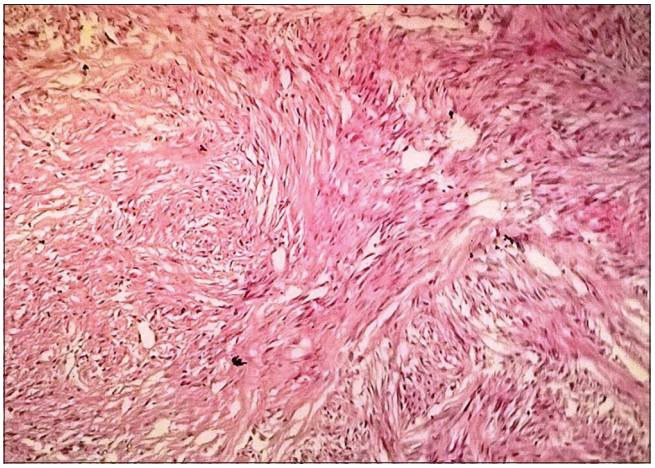

Figure 2: The tumor cells were spindle shaped with blunt-ended cigar shaped nuclei and abundant eosinophilic cytoplasm without cytological atypia (H and E, ×400)

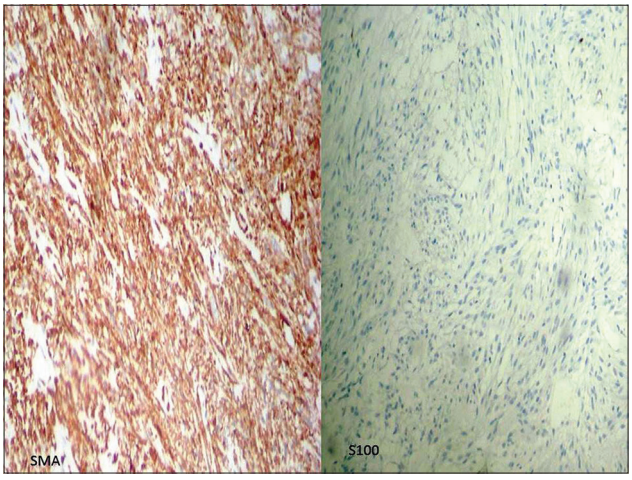

Figure 4: Immunohistochemistry study revealed positivity of smooth muscle actin and negativity of S100 protein

Solitary piloleiomyomas are infrequently painful, slightly larger than the multiple forms. Rarely they are present at birth and have slight female predominance.[6] Our patient had presented with 2 cm small solitary nodule on his left arm.

Solitary genital leiomyomas are usually asymptomatic and arise from the superficial smooth muscles of the scrotum, the labia majora or rarely nipples. Solitary angioleiomyomas are usually subcutaneous and rarely intracutaneous and are derived from vascular smooth muscles. The lower extremities are the most common sites and do not exceed 4 cm in diameter. Cutaneous angiolipoleiomyoma is a rare and an acquired asymptomatic tumor, always acral. They are more common in men and contrary to renal angiomyolipomas, have no association with tuberous sclerosis.[6]

The precise etiology and pathogenesis of this disease is still unknown. Apoptotic and antiapoptotic factors may play a role in the pathogenesis of leiomyomas. Wortham *et al*.[9] had reported an increase in antiapoptotic BCLX with a simultaneous decrease in proapoptotic BCL2 antagonist/killer protein.

Clinically patients present with either a solitary nodule or multiple firm dermal nodules located on the extremities and trunk.[10] Rarely they occur on the head and neck and easily mistaken for other cutaneous conditions. Hoyt *et al*.[11] reported solitary pilar leiomyoma of the nasal dorsum. Cutaneous leiomyomas are more common in adults than children, however, Dilek *et al*.[12] reported a case of cutaneous leiomyoma in a 10-year-old child who presented with multiple, red-brown masses on the back. Patients may complain of itching or pain. Pain may be triggered by pressure, cold temperature, trauma, or emotional stress. Pain may be caused by the pressure of the nerve fibers within the tumors upon contraction of the smooth muscles.[5]

Malhotra *et al*.[1] studied 37 cases of cutaneous leiomyoma in last 8 years. They reported 27 cases of piloleiomyoma, 3 cases of angioleiomyoma, 5 cases of breast leiomyomas and 2 cases of scrotal leiomyoma in patients ranging from 21 to 65 years of age. Solitary lesions were more common than multiple. The trunk and upper limbs were most commonly involved.

Raj *et al*.[13] reported cutaneous pilar leiomyoma: Clinicopathologic analysis of 53 lesions in 45 patients. They studied to analyze their clinicopathologic features. They noted 29 lesions from 21 patients of multiple tumors and 18 patients of solitary tumors.

Piloleiomyoma is the most common type of cutaneous leiomyoma and is unencapsulated, circumscribed dermal tumor composed of interlacing and whorled arrangement of smooth muscle fibers without atypia and mitotic activity. The nuclei are cigar shape with blunt ends. The differential diagnosis includes other spindle cells tumors such as neurofibroma, dermatofibroma, schwannoma, fibromyoma, and smooth muscle hamartoma. Histopathological examination, special stain, and immunohistochemistry may help to confirm the diagnosis of leiomyoma and differentiate them from other similar entities. In our case, we did special stain (MTS) and immunohistochemistry, which showed positivity for smooth muscle and confirmed the diagnosis. Excision is the only definitive treatment and recurrence after excision is rare.[4] Our patient was also treated by excision and has no recurrence in a follow-up period of 6 months.

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Nil.

Conflicts of interest
There are no conflicts of interest.

CONCLUSION

Cutaneous pilar leiomyoma is a rare benign smooth muscle neoplasm and is easily misdiagnosed for other cutaneous conditions. Hence, this lesion should always be taken into consideration in the clinical differential diagnosis of papulonodular skin lesions.

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