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Case report Open Access

A Rare Case of Zosteriform Pilar Leiomyoma

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Abstract

Background:

Cutaneous pilar leiomyoma is a benign, smooth muscle tumor, arising from erector pili muscle. The disease often affects large areas of body. In zosteriform leiomyoma, also called unilateral or segmental leiomyoma, lesions are distributed along a dermatome or extremity.

Main observations:

A 19 year old male patient had painful skin papules on external surface of right arm. One of the lesions was excised and leiomyoma was diagnosed histopathologically. Pain was relieved with topical nitroglycerin and lidocaine cream.

Conclusions:

Zosteriform pilar leiomyoma has cutaneous pilar leiomyoma's clinical and histopathological features. Although, in cutaneous pilar leiomyoma, typical lesions of disease spread over a wide area of body, it should be known that disease may be confronted with different distribution pattern. This case of zosteriform leiomyoma is presented because it is very rare.

Keywords: Zosteriform, segmental, pilar leiomyoma

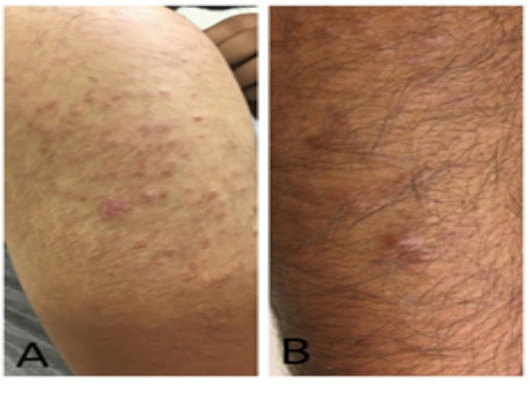
Introduction

Cutaneous leiomyoma is a rare benign smooth muscle tumor. It is divided into subtypes according to structure of origin. Cutaneous pilar leiomyoma develops from pili muscle, angioleiomyoma develops from vascular wall, and genital leiomyoma develops from scrotum, labia major, and nipple smooth muscle (1). Pilar leiomyoma is superficial lesion involving large area of body and is the most commonly found on extensor faces of extremities. Zosteriform leiomyoma, also called unilateral and segmental, is very rare (2). We aimed to present a case with zosteriform pilar leiomyoma in which pain can be controlled by topical calcium channel blocker and topical anesthetic treatment.

Case report

A 19-year-old male patient presented with a complaint of skin swelling on outer surface of right arm, causing severe pain during touching and showering (Figüre 1).

Figüre 1: Multiple, red-brown, painful lesions on outer surface of shoulder (A) and right arm (B).



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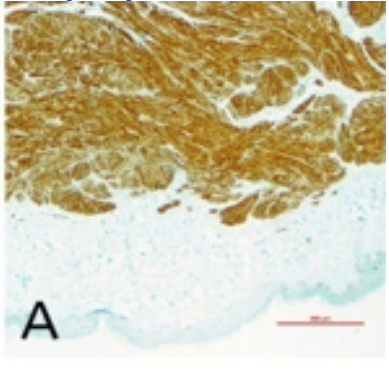
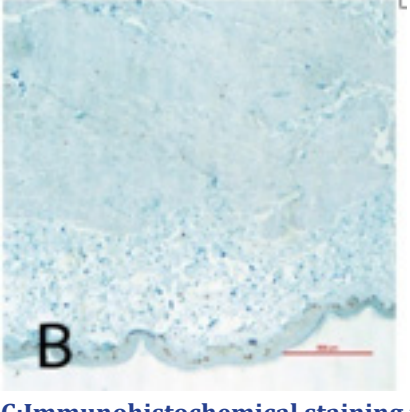
Patient's complaints have begun three years ago. Although number of lesions increased rapidly in first year, there was no new lesion for 2 years. Patient had no known disease and had no similar lesions in his line. Lesions were clustered on outer surface of right arm and on right shoulder. In affected area, there were pinkish brown, painful with touch, numerous papules and nodules. Patient had atrophic striae in same region. One of lesions was excised and histopathologically evaluated. Lesion was stained with hematoxylin eosin and intervening smooth muscle bundles were seen. When assessed immunohistochemically, cells stained strongly positive for smooth muscle actin (SMA) and demsin (Figüre 2).

Figüre 2: Histopathological images of the lesion.

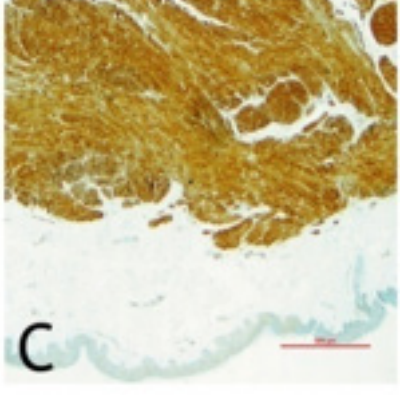
A: Immunohistochemical staining for smooth muscle actin (SMA).



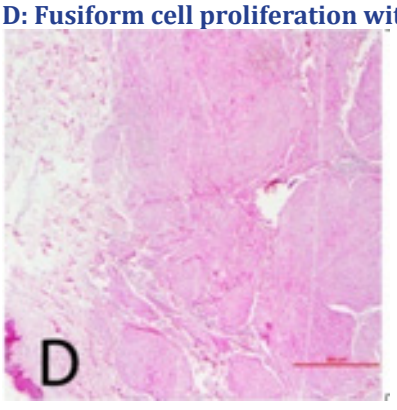
B: Kİ67 proliferation index is below 1%.



C:Immunohistochemical staining for desmin.



D: Fusiform cell proliferation with crossed bundles.



With diagnosis of pilar leiomyoma, nifedipine 3x10 mg capsule was started to patient. Patient, who used drug for 4 months, initially stated that despite pain relief, drug gradually lost its effectiveness. Patient applied to our clinic with a different treatment expectation. Botulinum toxin administration was recommended to patient. However, patient did not accept it because of its invasiveness, lack of permanent treatment and cost of treatment. Patient was started with nitroglycerin cream 2x1 and lidocaine hydrochloride pomade 2x1. At the end of first month, treatment satisfaction was provided.

Discussion

Cutaneous pilar leiomyoma is painful, red-brown, papulonodular-shaped, benign, smooth muscle tumors that develop in second and third decades of life. Most commonly found in face, back and extremities (3). Lesions may be solitary, but in most cases multiple. In our case, lesions were located on extensor face of right arm. In a study conducted by Ghanadan et al. 25 patients with cutaneous leiomyoma were evaluated. Although disease can be seen at any age, mean age of onset was 44 years. In same study, it was reported that 80% of cases were male and extremities were most common affected area (4). Lesions become obvious when physically stimulated by trauma or cold. Retained area contains numerous, 5-15 mm in size, reddish brown dermal nodules. In most cases of cutaneous leiomyoma, lesions are found in many parts of body. Rarely it can be seen in zosteriform pattern as in our case. Exact cause of Zosteriform patern could not be understood. It is thought to be related to presence of cutaneous nerves under retained region (2). Familial predisposition and association with visceral leiomyoma have been reported in multiple cutaneous

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leiomyomas. Leiomyomas can occur in every tissue containing smooth muscle. Visceral leiomyoma is common in retroperitoneal region and gastrointestinal tract. This case, which had no similar lesions in his family, had no esophageal or peptic complaints. Family history and visceral involvement are rare in Zosteriform pattern. However, in literature, a hysterectomy story was detected due to endometriosis in a patient. There are no cutaneous lesions in family of same patient, and a hysterectomy story related to uterine leiomyoma has been reported in her mother and cousin (5). Cutaneous leiomyomas are usually asymptomatic, contraction of tumoral muscle bundles and compression of nerve bundles may cause pain (6). Diagnosis is made by histopathologic examination and ice provocation test helps diagnosis. After ice cubes have been in contact with lesions for 15 seconds, a "goose flesh" appearance appears at skin. Severe pain occurs in contact area (2). Treatment of symptomatic solitary lesions is excision. However, a local recurrence of 50% may be seen and it is impractical to remove a large number of lesions. Successful use of various systemic and topical agents has been reported in literature. In systemic treatment, calcium channel blocker, nitroglycerin, antidepressant, etaverine and analgesic were used; in topical treatment, nitroglycerin, lidocaine, phentolamine, and hyoscine hydrobromide have been used and positive results have been reported (6). Botolinum toxin administration is another current treatment option and positive results have been reported in treatment of cutaneous leiomyoma (7). In this patient, pain relieved with topical nitroglycerin and lidocaine cream.

Conclusions

In conclusion, zosteriform pilar leiomyoma is a rare cutaneous leiomyoma. Zosteriform pilar leiomyoma has cutaneous pilar leiomyoma's clinical and histopathological features. With presentation of this case, it is desired to draw attention to distribution of zosteriform pilar leiomyoma.

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