Case Report

Cutaneous leiomyoma in a male: An uncommon case report

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ABSTRACT

Cutaneous leiomyomas, also known as pilar leiomyomas, arise from the arrectores pilorum muscles and are made up of a poorly circumscribed proliferation of haphazardly arranged smooth muscle fibers located in the dermis that appears to infiltrate the surrounding tissue and may extend into the subcutis. Cutaneous leiomyomas are rare, benign, smooth muscle tumors. The most common type of cutaneous leiomyoma, angioleiomyoma arise from the tunica media of blood vessels. The other subtypes, piloleiomyomas and genital leiomyomas arise from the arrector pili musculature of the hair follicle and from the smooth muscle found in the scrotum, labia, or nipple, respectively. Here, we presented a case of cutaneous leiomyoma, in a young male presenting with a swelling in the arm.

Key words: Dermal tumor, Leiomyoma, Smooth muscle

Leiomyomas are benign soft-tissue neoplasms arising from the smooth muscles. The skin is the second commonest location for leiomyoma after the uterus which accounts for 95% of cases. Cutaneous leiomyomas account for 75% of extra-uterine leiomyomas [1]. The World Health Organization defines cutaneous leiomyomas as benign dermal smooth muscle tumors derived from intrinsic dermal arrector pili smooth muscle or dartoic/vulvar/areolar smooth muscle. These are also known as piloleiomyoma [2]. Data regarding the prevalence and incidence of cutaneous leiomyomas are limited. Piloleiomyomas tend to occur much more frequently in adults than in children [3]. Pilar leiomyomas are rare and are more common in females than in males [2].

CASE REPORT

A 30-year-old male presented to the general surgery OPD with a swelling on the outer aspect of the arm (extensor surface) for the past 5 years. The patient also complained of pain occurring spontaneously or with provocation by pressure. The pain was typically described as dull aching in quality. A family history of similar lesions was not obtained in the patient at the time of presentation. No significant medical or surgical history was elicited.

General physical and systemic examination was unremarkable. On local examination, the mass was firm, tender, measuring about 13 cm in maximum dimension. The overlying skin showed multiple nodules.

Complete surgical excision was done and the specimen was received at the histopathology department. Gross examination revealed a skin covered tissue piece, reddish-brown in color, measuring 10 × 8 × 2 cm (Fig. 1).

Microscopically, the mass showed poorly circumscribed dermal nodules of intersecting well-differentiated smooth muscle bundles composed of spindle cells with copious brightly eosinophilic cytoplasm and elongated blunt-ended nuclei (Fig. 2). Immunohistochemically, the cells showed membrane expression of desmin and smooth muscle actin (Fig. 3). On the basis of histopathology and immunohistochemistry, it was diagnosed as cutaneous leiomyoma. However, follow-up of the patient could not be done.

DISCUSSION

Three distinct types of cutaneous leiomyomas such as piloleiomyomas, angioleiomyomas, and genital leiomyomas are described. This classification hinges on the origin of the smooth muscle tumor and corresponds to the histologic or anatomic site where the leiomyomas are found. Piloleiomyomas are believed to arise from the arrector pili muscle of the pilosebaceous unit, whereas, angioleiomyomas originate from the smooth muscles (tunica media) present within the walls of arteries and veins. Cutaneous leiomyomas are more common in adults than in children. However, isolated reports of cutaneous leiomyomas in children exist, including a solitary cutaneous leiomyoma on the heel of a neonate at birth [4].

Access this article online	
Received - 26 January 2021 Initial Review - 11 February 2021 Accepted - 16 February 2021	Quick Response code
DOI: 10.32677/IJCR.2021.v07.i02.008	

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Multiple piloleiomyomas are generally described in those aged 10–30 years, whereas, solitary piloleiomyomas usually appear later. The clinical differential diagnosis includes angiolipoma, glomus tumor, eccrine spiradenoma, neurofibroma, nevus, and lipoma.

Uterine smooth muscle tumors have been demonstrated to have estrogen receptor and progesterone receptor immunoreactivity. As a corollary, gonadotropin-releasing hormone analog therapy is useful in treating uterine smooth muscle tumors. However, no significant immunoreactivity for these receptors was noted in 15 cutaneous leiomyomas by McGinley *et al*. [5] and recently by Sen *et al*. [6]. These results point to a difference in the pathogenesis of uterine and extra-uterine tumors.

Cutaneous leiomyomas are tumors arising from the arrector pili muscles, which are superficial. They are rare lesions and form an important clinical differential diagnosis of painful papulonodules. Cutaneous leiomyomas account for 75% of extra-uterine leiomyomas. They comprise approximately 5% of all leiomyomas [1]. The extensor surfaces of the extremities are most commonly affected, followed by the trunk and the head-and-neck region. Genital leiomyomas can involve the scrotum, vulva, nipple, or areola. Multiple leiomyomas are more often painful. The distribution of multiple lesions was variable. Most commonly, grouped distribution was noted. Linear, segmental, and zosteriform variants have been described. Besides, zosteriform lesions which follow a dermatome, lesions following Blaschko's lines of fetal development, have been described [7]. Solitary leiomyomas are usually no larger than 2 cm. Genital leiomyomas are usually solitary. Vulvar tumors may enlarge during pregnancy.

Figure 1: Gross picture of cutaneous leiomyoma

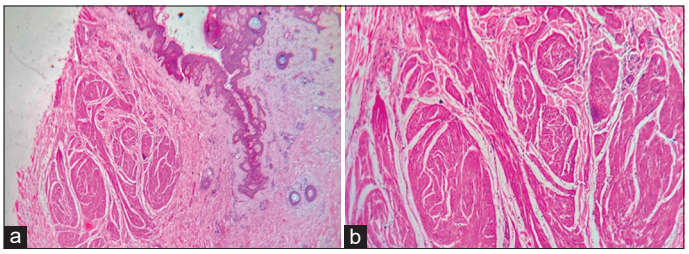

Figure 2: Microscopic picture of cutaneous leiomyoma (a) (H&E ×100); (b) (H&E ×400)

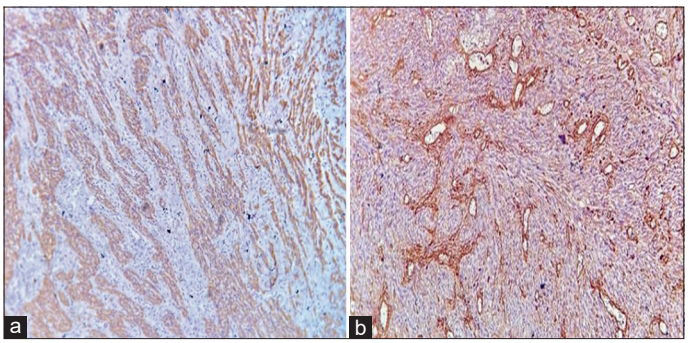

Figure 3: (a) Membranous expression of desmin (×100); (b) smooth muscle actin expression (×100)

The pathogenesis of pain associated with these lesions is unresolved. Some authors have suggested that pain could result from local pressure by the tumor on cutaneous nerves [8]. Others have hypothesized that muscle contraction may be central to the induction of pain [9]. The prominence of nerve fibers in angioleiomyomas is well described [10]. Symplastic leiomyomas akin to the uterine counterparts have been described in the skin [11]. SMA, desmin, and caldesmon are typically expressed.

Although theoretically the list of differential diagnoses is long, the use of Masson trichrome and desmin stains excludes most possibilities. Leiomyosarcoma is usually solitary and located on the lower extremity. Uterine smooth muscle tumors have been demonstrated to have estrogen receptor and progesterone receptor immunoreactivity.

CONCLUSION

We presented a rare case of cutaneous leiomyoma in a middle-aged male. These lesions must be biopsied to differentiate them from spindle cell lesions such as dermatofibroma, neurofibroma, and myofibroblastic lesions such as nodular fasciitis, fibromyoma, and smooth muscle hamartoma. Smooth muscle hyperplasia is a differential diagnosis of cutaneous leiomyoma, distinguished by its congenital or earlier onset, patch or plaque appearance, lack of associated tenderness, and more haphazardly arranged and discrete smooth muscle bundles. Combination of histopathological and immunohistochemical features aids in diagnosis.

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Funding: None; Conflicts of Interest: None Stated.

How to cite this article: Nandi S, Das C, Mukhopadhyay M, Ghatak A. Cutaneous leiomyoma in a male: An uncommon case report. Indian J Case Reports. 2021;7(2):64-66.