

Case Report

Salvage reconstruction for a case of leiomyoma after delayed diagnosis for twenty years

一例平滑肌瘤延遲診斷二十年後的搶救重建手術

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Correct clinical and histopathological correlation is important for the diagnosis of unusual clinical conditions. We report on a case of a 47-year-old woman patient who presented with multiple painful lesions over the right leg which had been treated with repeated steroid injections for over 20 years. Despite treatment, she continued to suffer from chronic pain and insomnia. Skin biopsy was eventually performed which showed pilar leiomyoma, which was treated with radical excision, and free flap reconstruction. The postoperative result was satisfactory with no recurrence at follow-up at one-year. Pilar leiomyoma is differentiated from multiple scars by correlation between the clinical history, physical examination and histopathology.

正確結合臨床和組織病理學相關性對於異常臨床狀況的診斷實為重要。我們報告了一例 47 歲女性患者，她右腿出現多處疼痛性病變，曾經重複接受類固醇注射治療達 20 多年之久。儘管接受了治療，她仍然受到慢性疼痛和失眠症的煎熬。患者最終進行了皮膚活檢，結果顯示為毛髮平滑肌瘤，並接受了根治性切除和游離皮瓣重建手術治療。術後效果滿意，隨訪一年未有復發。通過臨床病史、體格檢查和組織病理學之間的相關性結合，毛髮平滑肌瘤可以與多處癍痕區分開來。

Keywords: Connective tissue disorders, delayed diagnosis, surgery

關鍵詞：結締組織病、延遲診斷、手術

Introduction

Clinicopathological correlation is essential for the diagnosis of uncommon skin conditions. We present an unusual case of pilar leiomyoma which

was misdiagnosed and treated by different dermatologists as a cluster of scars for over 20 years. After the diagnosis was confirmed by skin biopsy, complete excision was done, followed by free flap reconstruction. There was no recurrence after surgery.

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Case report

A 47-year-old woman presented with multiple painful skin lesions on the right lower leg, which persisted despite multiple steroid injections for over

20 years. On examination, there were multiple bean-sized, skin-colored nodules over the anterior and medial sides of the right lower leg. The patient had been treated with monthly steroid injection every month for over 20 years. However, more lesions continued to appear and the pain was not relieved, which resulted in chronic insomnia. Excisional biopsy revealed a lesion that was composed of elongated spindle cells with blunt-ended nuclei consistent with pilar leiomyoma in the deep dermis and subcutaneous tissue. There was no evidence of malignancy. Therefore, she was referred to our clinic for further surgical intervention (Figure 1).

During the operation, the area with involvement by leiomyomata was 14x19 cm². This was excised down to the fascia and sent for frozen section. As the histopathological examination revealed benign lesions, complete excision was done. This was followed by reconstruction with a latissimus dorsi free flap as the extensor digitorum muscles and tibialis anterior muscles were exposed due to atrophy of subcutaneous tissue caused by repeated steroid injection. The remaining defect was covered with split-thickness skin-graft. After the operation, the patient made an uneventful recovery with no local recurrences at one-year of follow-up (Figure 2).

Discussion

According to the histological classification by the World Health Organisation, leiomyoma is divided into three different types: pilar leiomyoma, vascular leiomyoma and epithelioid leiomyoma.¹ Pilar leiomyoma is the most common type of leiomyoma, and arises from the erector pili muscle with histological expression of desmin (a muscle-specific protein). Pilar leiomyoma may present as a solitary tumor or multiple lesions, which are typically a group of red-brown papules.² The lesions are often painful, which may be exacerbated by cold exposure, pressure or emotional stress.³ With slow growth, leiomyomata persist for several years before any treatment is given. Malignant transformation may rarely occur. Due to the lack of specificity of clinical and sonographic findings, the diagnosis may only be made histopathologically.⁴

Due to its rarity, pilar leiomyoma may not be included in the differential diagnosis for subcutaneous lesions, as opposed to keloids and hypertrophic scars.⁵ Keloids may develop years after the initial injury, which can extend beyond original boundaries and become an irregular cluster of scars. On the contrary, hypertrophic scars occur within one to two months after injury, which generally remain confined to the

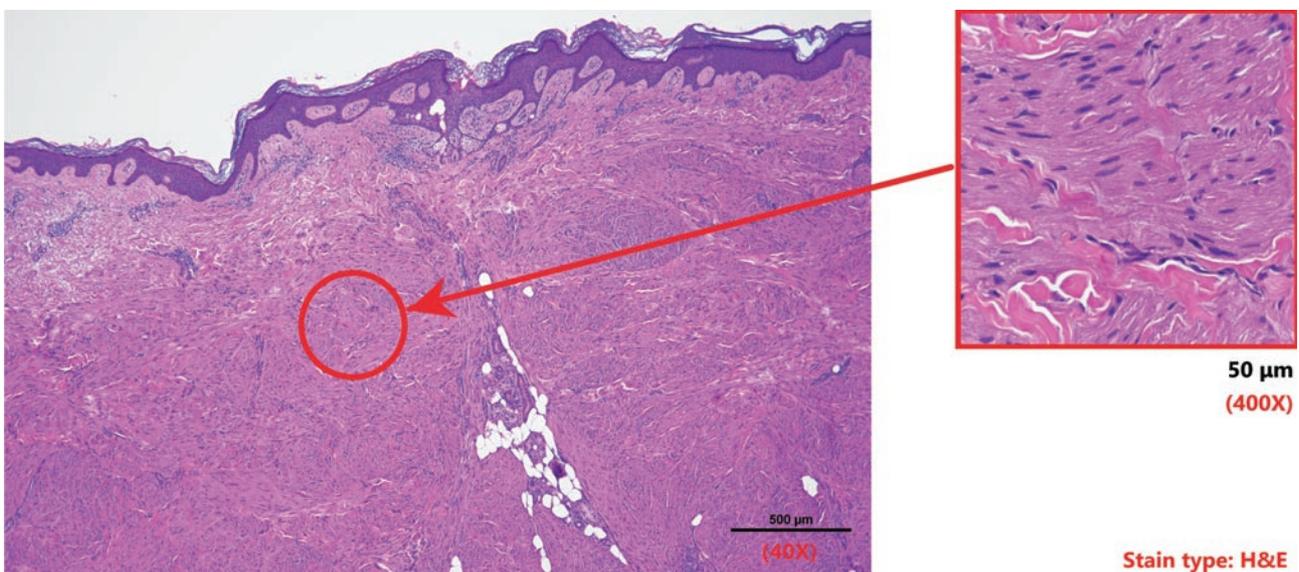


Figure 1. Histopathology of pilar leiomyoma (H&E stain x 40 and x 400). The lesion is composed of elongated spindle cells with blunt-ended nuclei.



Figure 2. (a) Before surgery, multiple satellite lesions were seen on the right leg; (b) Intra-operatively, the muscle tendons are exposed after excision of the tumour; (c) After surgery, no recurrence at one-year of follow-up.

original wound borders.⁶ In addition, the intensity and duration of the stimulation will determine the direction and growth speed of pathological scars. For instance, the horizontal tension direction on the anterior chest wall due to the pectoralis major muscle lead to extension of the scars in a horizontal direction.⁷ However, in our patient, the lesions of pilar leiomyoma spread out in a multiple satellite pattern, instead of the linear fashion of scars.

Among the various methods described, intralesional corticosteroid injection has been proven to induce the regression of keloid and hypertrophic scars by reducing collagen synthesis and fibroblast proliferation.⁸ However, skin atrophy is the most frequent side effect because it reduces the synthesis and induces degradation of extracellular matrix proteins in fibroblasts, which irreversibly leading to skin thinning and fragility.⁹ In our case, the patient had been treated with monthly triamcinolone acetonide injections for over 20 years, which resulted in the

exposure of the tendon of muscles after excision of the tumour due to atrophy of subcutaneous tissue.

Conclusion

Pilar leiomyoma is a rare benign soft tissue tumour that should be included in the differential diagnosis of multiple scar-like lesions that are unresponsive to repeated steroid injections. To avoid misdiagnosis and delayed treatment, a skin biopsy should be performed. Complete surgical excision is the treatment of choice for pilar leiomyoma, which can lead to total resolution of the symptoms with a very low risk of recurrence.

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