Surgical Alternative for Multiple Pilar Cutaneous Leiomyoma of the Lower Limb: Case Report and Literature Review

Alternativa cirúrgica para leiomioma cutâneo pilar múltiplo de membro inferior: Relato de caso e revisão da literatura

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Abstract

Pilar leiomyoma, or benign neoplasia of the arrector pili muscles, is an uncommon pathology, although it is the most frequent type of cutaneous leiomyoma. It occurs in young adults, affecting both sexes equally. It presents as multiple or isolated, reddish-brown, papulonodular lesions. Although no malignant tumors have been reported, it is important to include this pathology in the differential diagnosis of papulonodular cutaneous lesions. Surgical treatment is often reserved for prolonged and symptomatic cases, with relapse reports. In this paper, we report the case of a 28-year-old male patient with a diagnosis of pilar leiomyoma on the lateral thigh and right leg who underwent a tangential skin resection.

Keywords

- plastic surgery
- leiomyoma
- lesion
- neoplasias
- skin

Resumo

O leiomioma pilar, ou neoplasia benigna do músculo eretor do pelo, é uma patologia incomum, apesar de ser o tipo mais frequente de leiomioma cutâneo. Ocorre entre adultos jovens, acometendo igualmente ambos os sexos. Sua apresentação se dá em lesões dolorosas, isoladas ou múltiplas, papulonodulares vermelhas e castanhas. Apesar de não ter sido relatada nenhuma malignização dos tumores, é importante incluir essa patologia no diagnóstico diferencial de lesões cutâneas papulonodulares. O tratamento cirúrgico geralmente é reservado a casos extensos e sintomáticos, com relatos de recidiva. Nesse artigo, relatamos o caso de um paciente de 28 anos, do sexo masculino, com diagnóstico de leiomioma pilar em face lateral de coxa e perna direita submetido à ressecção de pele tangencial.

Palavras-chave

- cirurgia plástica
- leiomioma
- lesão
- neoplasias
- pele

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Introduction

Leiomyomas are benign soft tissue neoplasms arising from smooth muscle. Cutaneous pilar leiomyoma is a rare benign tumor, with an incidence of approximately 5%, and can present in solitary or multiple forms (Ragsdale, 2009). Solitary lesions are more common than multiple ones (Malhotra, 2010).

This disease is more common in adults, although there are reported cases in children. They do not have a gender predilection, although data are conflicting in the literature (Raj, 1997).

The differential diagnosis includes neurofibroma, dermatofibroma, schwannoma, fibromyoma, and smooth muscle cell hamartoma (Latoni, 2000), especially in solitary lesions. Biopsy is the test used for diagnosis confirmation.

These lesions are usually refractory to analgesic treatment. Some reports suggest the efficacy of calcium channel blocker medications (such as nifedipine), alpha-blockers (such as phenoxybenzamine), nitrates, and gabapentin (Raj, 1997). A patient with clinical contraindications for surgery underwent CO_2 laser ablation with good outcomes.

Objective

We herein report the case of a 28-year-old male patient with multiple cutaneous leiomyomas who underwent surgical therapy.

Case Report

A 28-year-old male patient of mixed race presented with multiple reddish-brown papular lesions of varying sizes, isolated in some regions, and coalesced in parts of the right thigh and leg. The lesions appeared in the last 5 years. The patient presented with pain, mainly when exposed to cold, which limited his movement and work activity. The pathological diagnosis confirmation of pilar leiomyoma relied on two biopsies. It was not feasible to resect the lesions separately. The patient underwent surgery at Hospital Público Regional de Sobradinho, Brasília, DF, Brazil, on April 18, 2022. This research was approved by the Ethics in Research Committee under the protocol number 69524923.0.0000.0257.

We opted for tangential resection with a Blair knife in the thigh area with the highest density of lesions, measuring 14 by 8 cm on the anterior surface (►**Fig. 1**). At 8 months after the procedure, the patient reported pain improvement in the operated area, with no keloid scar formation (►**Fig. 2**). For the anterolateral area of the thigh and right leg, we chose a wide and tangential resection with a Blair knife (►**Fig. 3**). The patient had no postoperative complications. In the first 6 months, he reported improvement in pain and quality of life. However, some lesions recurred in the operated area.

Discussion

Superficial (cutaneous) leiomyomas are benign smooth muscle tumors that may originate from the arrector pili muscles, dartos, vulvar, and mammary smooth muscles, or the smooth muscles surrounding the dermal blood vessels. Their classification includes piloleiomyomas (solitary or multiple), external genitalia leiomyomas, and angioleiomyomas (Kumar, Abbas, and Fausto, 2005). The first report of a cutaneous leiomyoma dates back to 1854 by Rudolf Virchow.

These are relatively uncommon neoplasms, and their exact incidence is unknown. The pathogenesis remains to

Fig. 1 Preoperative period.

Fig. 2 Postoperative period.

Fig. 3 Broad and tangential resection of the affected area.

be determined, but it is believed that piloleiomyomas arise from smooth cells within the arrector pili muscle of the pilosebaceous unit (Hoyt, 2015). Multiple piloleiomyomas may occur sporadically or be autosomal dominant inherited (with variable penetrance), as part of the Reed syndrome.

Piloleiomyomas, or pilar leiomyomas, may be solitary or multiple. A single patient may present hundreds of lesions. They are firm, reddish-brown to skin-colored nodules or papules (Bologna, 2010). Solitary piloleiomyomas develop mainly during adulthood, with a similar gender distribution. When multiple, the distribution pattern is most commonly clustered and linear following the Blaschko lines, but scattered lesions may occur. Most are 1 to 2 cm in diameter and usually develop on the extremities and trunk (especially the shoulder). Solitary lesions favor the limbs, while multiple ones mostly appear on the trunk (Cizmeci, 2007). They are often associated with spontaneous or induced pain, such as cold exposure.

Histologically, pilar leiomyomas are smooth muscle cell tumors intertwined and arranged in a spiral, without atypia or mitotic activity.

Surgical excision is the indicated treatment for cases with few lesions and may be curative for solitary or limited tumors. However, 50% of multiple lesion cases recur within 6 months to 15 years. For numerous and painful piloleiomyomas, some authors recommend a drug treatment, despite its limited efficacy.[11] Drug treatment includes gabapentin, oral or topical nitroglycerin, lidocaine, nifedipine, verapamil, phenoxybenzamine, phentolamine, hyoscine, analgesics, and antidepressants. Cryotherapy and electrocoagulation have shown little benefit (Suzuki, 2007).

Tangential resection is frequent in plastic surgery for wound and burn debridement or skin graft removal (Bolgiani and Serra, 2010). Recurrence is probably more common in lesions in the deeper dermis, with no tangential resection. In these cases, we suggest a new, more delimited tangential resection, potentially complemented by a CO_2 laser.

Conclusion

The literature does not report tangential excision for pilar leiomyoma treatment. The use of this technique in the case herein reported was efficient for multiple lesion treatment, especially for pain relief. Combined treatments may be effective in preventing recurrence and ensuring a cure with no complications.

Author's Contributions

IMS: analysis and/or interpretation of data, validation, and writing – original draft; ASC: supervision; LAA: formal analysis; LVD, methodology; MSV, data curation; and SFAG: writing – original draft.

Clinical Trial

None.

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Conflict of Interests

The authors have no conflict of interests to declare.

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