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LETTER TO THE EDITOR

Ear lobe angioleiomyoma and aggressive keloid

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Sir,

Angioleiomyoma is a rare, benign smooth muscle tumor originating from the tunica media of blood vessels. It may be localized anywhere in the body. Ear lobe angioleiomyoma is rarely diagnosed before surgery. The usual treatment is surgical excision. Ear lobe keloids are very common. Often, this entity comes as the result of ear piercing. The question of coexistence of an angioleiomyoma with an aggressive keloid is unusual. In February 2008, a 12-year-old female presented a nodular lesion of the right ear lobe. The patient referred previous ear piercing history at both earlobes without familiarity for keloids. A similar lesion had been excised at the same site in 2001. The histological diagnosis was "vascular leiomyoma." The lesion recurred in April 2003; a second excision was performed and the histology was the same as the first one. Between April 2003 and February 2008, a new lesion appeared at the same site. In February 2008, it was oval-shaped, with well-defined margins, and painless. This new local recurrence was excised with 0.3 cm clear margins and a postauricular flap was performed for right ear lobe reconstruction. Histology showed "remarkable nodular fibrosis with keloid-like aspects at the periphery; lesion completely excised." In May 2009, a new recurrence occurred at the junction between the new ear lobe and preauricular region (Fig. 1).

A second patient, 24 years old female, in December 2011 presented a lesion at the left ear lobe site. A nodular lesion was first noted in August 2007, adjacent to the site of a previous ear piercing. The lesion was round-shaped and painless. After its first excision (2007), the histology was "cutaneous leiomyoma incompletely excised; the inferior margin of the excision is still involved." In December 2007, she presented a recurrence and a second excision was performed; histology reported "cutaneous angioleiomyoma" again, with "unclear deep excision margins." In September 2010, a new recurrence appeared at the scar site. A wide surgical excision was performed again; histology reported "hypertrophic scar tissue with keloid focal features." All the procedures were performed by the same general surgeon. In September 2011, the scar became hypertrophic, then she came to our clinic. We applied the same protocol for both patients consisting in intralesional injections of triamcinolone acetonide 40 mg/ml diluted 1:1 in saline solution and "U-shaped" Zimmer splint ear lobe compression (Fig. 2).

The incidence range of angioleiomyoma is between the third and sixth decade of life, with a female prevalence. The lesion is usually a unique lesion, small, round, firm, skin-colored, and well-encapsulated [1, 2]. On the other hand, ear lobe keloid is very common. Often, this entity comes as the result of ear piercing. The exact pathogenesis of keloids is unknown. Most important factors involved in keloid formation are chronic inflammation, infection, excessive wound tension, foreign material, or other impurities in earrings [3, 4].

Many options have been proposed for keloid treatment: simple excision, intralesional injection of corticosteroids, application of pressure, and application of silicone gel or patches. The occurrence of an angioleiomyoma that ends into an aggressive keloid is extremely unusual. Luo and Pan [5] studied leiomyomas, keloids, surgical scars, and peritoneal adhesions to identify genes that could individually distinguish these fibrotic disorders despite differences in the nature of their development and growth. Leiomyomas in Caucasians compared with keloids in African-Americans showed a limited difference in their gene expression profile. This suggested the existence of a comparable environment for leiomyomas and keloids. Their results indicated that the molecular features

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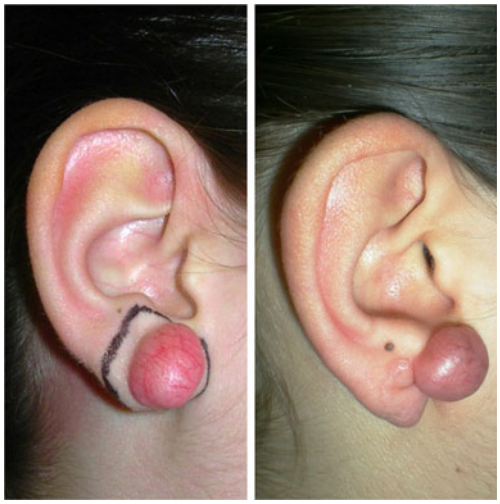

Fig. 1 First patient: Clinical recurrence of right ear vascular leiomyoma and its evolution into clinical ear lobe keloid after postauricular flap

of leiomyomas were comparable but might be under different tissue-specific regulatory control to those of keloids; furthermore, they differed in the tissue-specific expression of a selected number of genes regulating cell growth and apoptosis, inflammation, angiogenesis, and tissue turnover. Catherino et al. [6] demonstrated that decreased expression of dermatopontin was associated with keloid formation, a fibrotic disease that shared epidemiologic similarities with uterine leiomyoma. Immunohistochemical studies of leiomyomas and keloids showed reduced levels of dermatopontin in both tissues, and ultrastructural analysis revealed similar orientation of collagen fibrils in both entities despite the fact that they arose from different progenitor tissues. Molecular and immunohistochemical studies showed a correlation between the two entities. Therefore, we suggest, in case of nodular lesions at the ear lobe site, a careful clinical evaluation and a mindful treatment plan including lesion biopsy. In case of histological diagnosis of angioleiomyoma, we recommend surgical excision and immediate postoperative compression. It is possible that the two conditions arise from the patient's susceptibility to both without necessarily transforming from one entity to another. Future studies with genes profiling could be more convincing to identify and demonstrate a mutation common to the two entities.

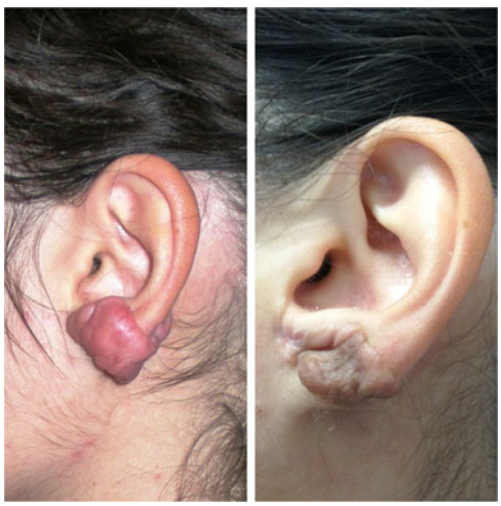

Fig. 2 Second patient: keloid on the left ear lobe after three surgical excisions and its involution after our treatment protocol

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