

## A rare case of intraoral trichilemmal cyst

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### **ABSTRACT:**

Trichilemmal cysts, also known as pilar cysts, are slow-growing lesions commonly found on the scalp of elderly women. They arise from outer root sheath epithelium and have a distinctive pattern of keratinization called trichilemmal keratinization. Here, we describe a rare case of a trichilemmal cyst on the lower lip mucosa of a 29-year-old man. The infrequency of intraoral hairs unquestionably results in the shortage of reported cases of trichilemmal cysts into the oral cavity.

**Keywords:** intraoral hair, pilar cyst, trichilemmal cyst.

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## INTRODUCTION

Trichilemmal cysts (TCs), also known as isthmus-catagen cysts or pilar cysts, are sometimes clinically indistinguishable from epidermal cysts. These cysts occur as solitary or multiple intradermal or subcutaneous lesions on the scalp of elderly women<sup>1</sup>. In general, they arise from outer root sheath epithelium<sup>2</sup>. Here, we describe a rare case of a trichilemmal cyst on the oral mucosa of a young man.

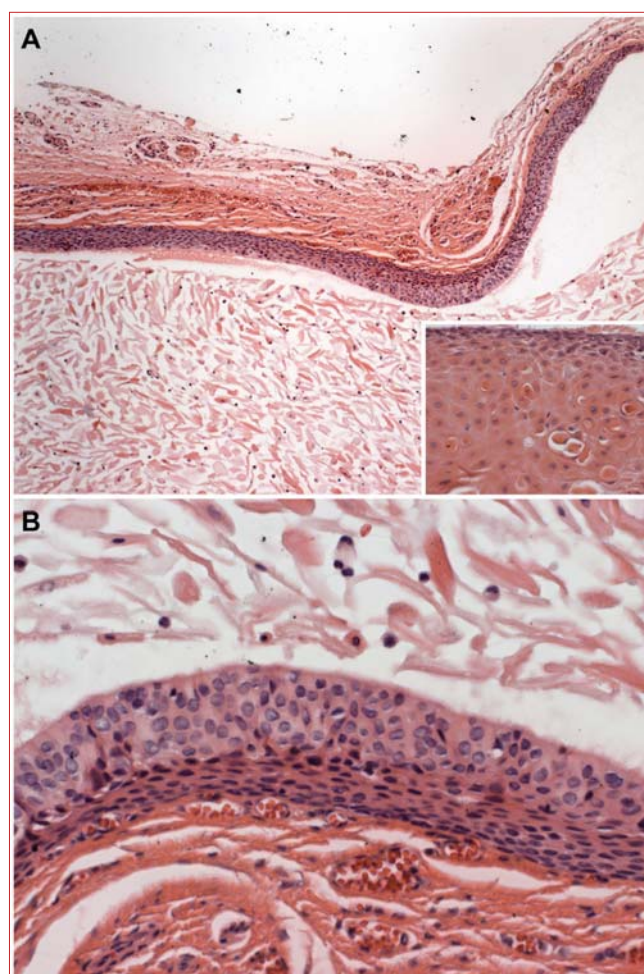
## CASE REPORT

A 29-year-old black man with a chief complaint of a lower lip mucosal swelling was referred to the Stomatology Service of the School of Dentistry, University of São Paulo. He had noticed the lesion 5 months earlier, and since then, no changes were observed. Physical examination revealed an asymptomatic, nodular soft lesion covered by normal mucosa and measuring 10 mm (Figure 1). The lesion was completely excised.



**Figure 1.** Intra-oral aspect of lower lip lesion. A nodular lesion recovered by normal oral mucosa.

Macroscopic analysis showed a brownish-yellow nodule measuring 20 × 10 × 10 mm and a well-established cavity filled with a yellowish-white dense mass. Histopathology revealed a lesion covered by a well-defined capsule comprising thin squamous epithelium. The basal cell layer was composed of cuboidal cells with hyperchromatic nuclei in a distinct palisade arrangement. The upper cell layer showed swollen cells with pale cytoplasm. An interesting feature was abrupt keratinization of epithelial cells without the formation of the granular layer. Mononuclear cells and eosinophilic material representing keratinous debris filled the cyst lumen. The cyst wall was composed of fibrous connective tissue with scattered chronic inflammatory cells (Figure 2). These histological findings determined the final diagnosis of trichilemmal cyst.



**Figure 2.** Histopathological aspects. A. A well circumscribed capsule delimited by thin squamous epithelium (original magnification 10x). In detail observe a basal cell layer of cuboidal cells with hyperchromatic nuclei and palisade arrangement and swollen cells with pale cytoplasm in the upper cell layer (original magnification 40x). B. Abrupt keratinization of the epithelial cells without the formation of an intervening granular layer and cyst lumen was filled by mononuclear cells and an eosinophilic material represented by keratinous debris (original magnification: 20x).

## DISCUSSION

Clinically, TCs differs from epidermal cysts in frequency and distribution: they are less common than epidermal cysts and about 90% of the TCs occur on the scalp<sup>1</sup>. Furthermore, in contrast to epidermal cysts, TCs appear as firm, smooth, white-walled cysts and are easily enucleated<sup>3</sup>. Epidermal cysts are relatively common in the oral cavity and usually found in the floor of the mouth. To the best of our knowledge, the current literature includes only one report of an intraoral TC<sup>4</sup>.

TCs typically present as sharply circumscribed cysts with keratinized cells<sup>5,6</sup>. The most striking finding that distinguishes epidermal cysts from TCs is the keratinization pattern: the presence or absence of keratohyaline granules within the cyst wall, namely epidermal keratinization or trichilemmal keratinization,

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is specific to this pathology<sup>7</sup>. In this pattern, cells undergo an abrupt transition from the stratum spinosum to the keratinized layer without the formation of the granular layer<sup>5,6</sup>. The peripheral layer of cells also shows a distinct palisade arrangement not seen in epidermal cysts<sup>8</sup>. Likewise, the surface epithelium of TCs is composed of large eosinophilic keratinocytes<sup>9,10</sup> and the lumen is filled with keratinous debris<sup>6</sup>.

A TC can transform into a proliferating trichilemmal tumor (PTT), which is usually observed as large lobulated scalp masses<sup>11</sup>. Both lesions share similar histological features, but extensive epithelial proliferation, variable cytological atypia, and mitotic activity are observable only in PTT<sup>2</sup>. In some instances, malignant transformation of PTT occurs, and this malignant lesion presents some areas reminiscent of trichilemmal keratinization but shows severe atypia and invasion of the surrounding tissue<sup>12</sup>.

Despite its rarity and unusual location, all histological features observed in the case led to the final diagnoses of TC. As it arises from outer root sheath epithelium, its occurrence in the oral cavity is rare; its formation can be explained by the presence of ectopic hair follicles in some oral sites, especially the lips. Moreover, cysts arising from deep hair follicles in the lip area can grow toward the mouth instead of the skin and present as oral lesions rather than skin lesions.

The hypothesis of PTT was discarded once the lesion was found to present the typical histological features of TC. Because TCs are usually well circumscribed, complete excision of the lesion was performed. The patient is under periodic follow-up. One year after the excision, no clinical evidence of recurrence was observed.

Considering the relatively low incidence of TCs in the oral cavity, differential diagnosis with other similar lesions is required.

## REFERENCES

1. Elder DE, Elenitsas R, Johnson BL, Murphy GF. *Lever's histopathology of the skin*. Philadelphia: Lippicott Williams & Wilkins; 2005.
2. Folpe AL, Reizenauer AK, Mentzel T, Rutten A, Solomon AR. Proliferating trichilemmal tumors: clinicopathologic evaluation is a guide to biologic behavior. *J CutanPathol*. 2003 Sep;30(8):492-8.
3. Leppard BJ, Sabderon KV, Wells RS. Hereditary trichilemmal cysts. *ClinExpDermatol*. 1977 Mar;2(1):23-32.
4. Perez LM, Bruce JW, Murrah VA. Trichilemmal cyst of the upper lip. *Oral Surg Oral Med Oral Pathol Oral RadiolEndod*. 1997 Jul;84(1):58-60.
5. Sau P, Graham JH, Helwig EB. Proliferating epithelial cysts: clinicopathological analysis of 96 cases. *J CutanPathol*. 1995 Oct;22(5):394-406.
6. Shet T, Rege J, Naik L. Cytodiagnosis of simple and proliferating trichilemmal cysts. *ActaCytologica*. 2001;45(4):582-8.
7. Ohnishi T, Watanabe S. Immunohistochemical observation of cytokeratins in keratinous cysts including plantar epidermoid cyst. *J CutanPathol*. 1999;26(9):424.
8. McGavran MH, Binnington B. Keratinous cysts of the skin: identification and differentiation of pilar cysts from epidermal cysts. *Arch Dermatol*. 1966;94(4):499-508.
9. Ackerman AB, Viragh PA, Chongchitnant N. *Neoplasms with follicular differentiation*. Philadelphia: Lee & Febiger; 1993.
10. Shimizu Y, Sakita K, Arai E, Tsuchida T, Ogawa F, Ban S, et al. Clinicopathologic features of epidermal cysts of the sole: comparison with traditional epidermal cysts and trichilemmal cysts. *J CutanPathol*. 2005;32(4):280-5.
11. Chang SJ, Sims J, Murtagh FR, McCaffrey JC, Messina JL. Proliferating Trichilemmal Cysts of the Scalp on CT. *Am J Neuroradiol*. 2006 Mar;27(3):712-14.
12. Mehregan AH, Lee KC. Malignant proliferating trichilemmal tumors: report of three cases. *J DermatolSurgOncol*. 1987;13(12):1339-42.