ABSTRACT – Trichilemmal horn or trichilemmal keratinosis is a benign neoplasm, that occurs more frequently in females and in the age group over 50 years old. Clinically, it presents as a cutaneous horn and histologically shows trichilemmal keratinization at the base of the lesion. As an uncommon pathology, in this case report, we will present the case of a young man (14 years old), presenting a keratotic lesion on the forehead for eight months, which revealed the diagnosis of trichilemmal horn after lesion exeresis and anatomopathological analysis.

KEYWORDS – Forehead; Keratosis.

RESUMO – O corno triquilemal ou ceratose triquilemal é uma neoplasia benigna, que ocorre com maior frequência no sexo feminino e na faixa etária acima de 50 anos. Clinicamente, apresenta-se como um corno cutâneo e histologicamente mostra ceratinização triquilemal na base da lesão. Como patologia incomum, neste relato de caso, apresentaremos o caso de um jovem (14 anos), apresentando lesão ceratótica na fronte com oito meses de evolução, cujo estudo anatomopatológico revelou o diagnóstico de corno triquilemal após exérese da lesão.

PALAVRAS-CHAVE – Queratose; Testa.

INTRODUCTION

Trichilemmal horn or trichilemmal keratinization is considered a benign and very rare neoplasm,\(^1\) characterized in the histopathology by an abrupt maturation of keratinocytes in lamellar keratin without the formation of the granular layer, occurring in the outer sheath of the hair follicle root and in the trichilemmal sac of the catagen and telogen wires.\(^2\) The prevalence is higher in women and in the age groups older than 50 years, with a predominance in extremities followed by the cephalic region.\(^1-3\) Its etiopathogenesis remains uncertain,\(^2\) however there are reports of its occurrence secondary to the rupture of a previous trichilemmal cyst.\(^3\) The diagnosis is confirmed after excision and histopathological analysis, showing specific characteristics of this pathology.

CASE REPORT

A 14-years-old male, attended the dermatology clinic at Heliópolis Hospital Complex, São Paulo, referring the appearance eight months before of an asymptomatic papular lesion on the forehead without association with trauma or local procedures. He reported progressive growth with no change in the characteristics of the lesion.

On dermatological examination, there was a well-defined papule with an exophytic keratotic center, measuring approximately 1 cm (Fig. 1). Initially, keratoacanthoma, traumatized verruca vulgaris and cutaneous horn were considered as possible diagnoses. After surgical excision the anatomopathological analysis stained by the hematoxylin-eosin (HE) showed a well-limited lesion where the epidermis had thick hyperkeratosis and papillomatosis, with compact keratinization without the presence of a granular layer. The more superficial keratinocytes, unlike what would be expected in the more superficial layers, had bulky and vesicular nuclei (Fig.s 2,3 and 4), but no atypia, which is consistent with the diagnosis of a trichilemmal horn. In the dermis, vascular ectasias and fibrosis are observed with no significant inflammatory infiltrate.

The patient returned 45 days after the excision for reassessment, showing complete resolution of the condition.

DISCUSSION

Trichilemmal horn is an uncommon lesion with a unique histology, characterized by an area of papilomatous epidermis with the
absence of the granular zone, giving rise to a compact, eosinophilic and orthokeratotic keratin, thus characterizing trichilemmal keratinization. There may be nodular extension to the dermis, but inflammation in this location is usually absent,\(^1,2\) as in the present case.

The etiopathogenesis remains uncertain and the participation of the human papilloma virus (HPV) in this process has been suggested, as previous microscopy studies showed intranuclear particles with morphology similar to HPV, however immunohistochemical studies with anti-HPV antibodies were negative.\(^1\) The CD34 antigen is a specific marker for external sheath of the hair follicle and tumors derived or differentiated for this type of epithelium are positive, as in the cases described as trichilemmal horn.\(^2,3\)

Some cases reported the appearance of trichilemal horn after rupture of trichilemal cysts,\(^2\) which was not the case of our patient.

Macroscopically, the lesion is characterized by an exophytic and keratotic lesion, measuring approximately 1 to 2 cm in diameter.\(^1\) The prevalence is higher in women and in the age group after 50 years, however cases have been described in ages ranging from 16 to 72 years. Lower limbs, followed by the head and back are the most affected regions.\(^1,2\) As there is a higher prevalence in body extremities and cephalic region (photoexposed areas), the direct or indirect contribution of ultraviolet (UV) radiation to the disease pathogenesis in some patients has been discussed.\(^2\)

The diagnosis is made through macroscopic analysis and microscopic examination of the lesion, through excision and anatomopathological analysis.\(^2\) Possible differential histological diagnoses of the trichilemmal horn are hypertrophic actinic
keratosis, keratoacanthoma, seborrheic keratosis, trichilemal cyst, trichilemoma and verruca vulgaris, among others.

In the case of the aforementioned patient, both the macroscopic lesion and the anatomopathological characteristics were consistent with the changes described in trichilemmal horns, as well as the location of the lesion, however it diverged from the literature data in relation to age (14 years) and sex.

We decided to report this case as the trichilemmal horn is an uncommon lesion, having as differential diagnosis frequent lesions in the daily dermatological routine and there is very little data in literature about this pathology.

Conflicts of Interest: The authors have no conflicts of interest to declare. Financing Support: This work has not received any contribution, grant or scholarship. Confidentiality of Data: The authors declare that they have followed the protocols of their work center on the publication of data from patients. Patient Consent: Consent for publication was obtained. Provenance and Peer Review: Not commissioned; externally peer reviewed.

REFERENCES
1. Haro R, González-Guerra E, Fariña MC, Martín-Moreno L, Requena L. Cuerno tricolémico: presentación de un nuevo caso y revisión de la literatura. Actas Dermosifiliogr. 2009;100:65-8.
2. DiMaio DJ, Cohen PR. Trichilemmal horn: case presentation and literature review. J Am Acad Dermatol. 1998;39:368-71. doi: 10.1016/s0190-9622(98)70393-7.
3. Iro S, Maadane A, Slimani F. Facial cutaneous horn in three different conditions: A highlight of a malignancy risk. Int J Surg Case Rep. 2021;83:105999. doi: 10.1016/j.ijsr.2021.105999.
4. Peteiro MC, Toribio J, Castelo JL. Trichilemmal horn. J Cutan Pathol. 1984;11:326-8. doi: 10.1111/j.1600-0560.1984.tb00384.x.
5. Nakamura K. Two cases of trichilemmal-like horn. Arch Dermatol. 1984;120:386-7.