

CASE REPORT

Trichilemmal Horn: a New Case and Review of the Literature

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Abstract. Trichilemmal horn, or trichilemmal keratosis, is an uncommon benign neoplasm of follicular lineage with trichilemmal differentiation.

The essential characteristics of this tumor are its clinical presentation in the form of a cutaneous horn with trichilemmal keratinization apparent in the histology study (with a hyperplastic epithelium giving rise to dense, orthokeratotic eosinophilic keratin).

We present a new case of this type of tumor in an 82-year-old woman who developed a solitary lesion on her scalp after surgical removal of a trichilemmal cyst. On the basis of the pathology report, the tumor was diagnosed as trichilemmal horn.

We review the 33 reports of this tumor in the literature to date.

Key words: trichilemmal horn, trichilemmal keratosis, follicular tumor, trichilemmal keratinization.

CUERNO TRICOLÉMICO: PRESENTACIÓN DE UN NUEVO CASO Y REVISIÓN DE LA LITERATURA

Resumen. El cuerno tricolémico o queratosis tricolemal es una neoplasia benigna, poco frecuente, de estirpe folicular y diferenciación tricolémica.

Las características fundamentales de este tumor son su presentación clínica en forma de cuerno cutáneo que histológicamente muestra queratinización tricolémica (con un epitelio hiperplásico que da lugar a una queratina eosinófila, compacta y ortoqueratósica).

En este artículo presentamos un nuevo caso de este tumor en una paciente de 82 años que desarrolló una lesión solitaria en el cuero cabelludo, tras la manipulación de un quiste tricolémico. El estudio histológico permitió llegar al diagnóstico de cuerno tricolémico.

Realizamos una revisión de los 33 ejemplos de este tumor descritos en la literatura hasta la fecha.

Palabras clave: cuerno tricolémico, queratosis tricolemal, tumor folicular, queratinización tricolémica.

Introduction

Trichilemmal horn, or trichilemmal keratosis, is a rare benign neoplasm with trichilemmal differentiation.

The essential characteristics of this tumor are its clinical presentation in the form of a cutaneous horn and its histological features, which show trichilemmal keratinization at the base of the lesion.¹

Thirty-three cases of trichilemmal horn have been described in the English-language literature.¹

Case Description

An 82-year-old woman with no relevant personal history presented to our hospital's dermatology department with an asymptomatic, rapidly growing lesion in the occipital region of the scalp, present for several weeks. The patient had several trichilemmal cysts on her scalp and reported that before developing this new lesion she had had a cyst in the same location that had been drained surgically.

Physical examination revealed a tumor in the form of a cutaneous horn, with a hyperkeratotic center of about 2 cm in diameter at the base and 1 cm in length, located on a fleshy erythematous base (Figure 1). On the basis of these

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Figure 1. Trichilemmal horn: papule with a hyperkeratotic center located on a fleshy erythematous base..

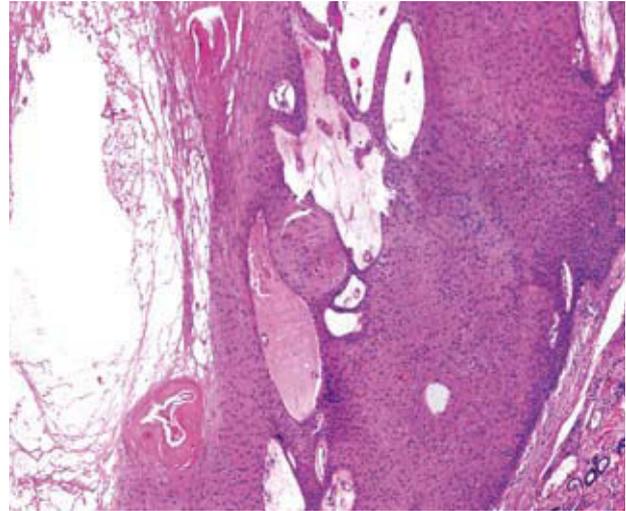


Figure 3. Detail of the lesion: epithelium with a peripheral layer of palisaded cuboidal cells lying on a basement membrane and several overlying layers of large squamous cells with abundant cytoplasm Hematoxylin–eosin, original magnification $\times 40$.



Figure 2. Crateriform lesion with a keratotic center and a base composed of a hyperplastic squamous epithelium showing trichilemmal keratinization. Hematoxylin–eosin, original magnification $\times 10$..

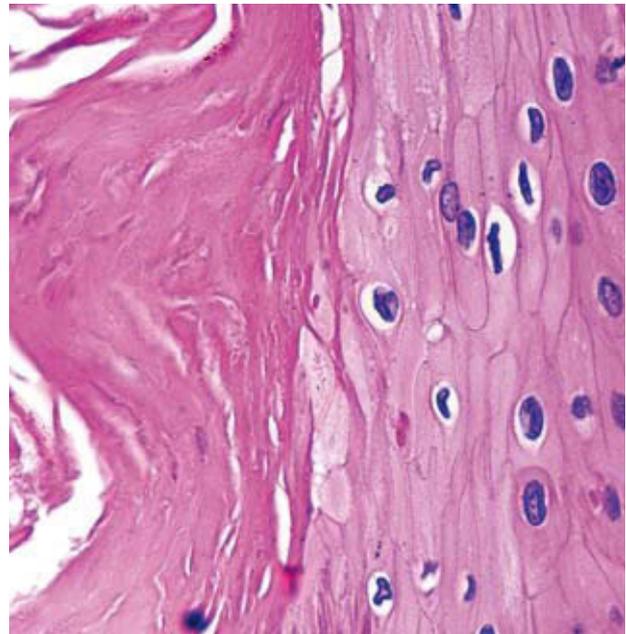


Figure 4. Detail of Figure 3. Keratinization with no granular layer, giving rise to dense orthokeratotic eosinophilic keratin. Hematoxylin–eosin, original magnification $\times 200$.

findings, a clinical diagnosis of keratoacanthoma was made and the lesion removed.

Histopathology revealed a well-delimited crateriform lesion with a keratotic center extending to the external surface of the skin and a base with a hyperplastic squamous epithelium showing trichilemmal keratinization (Figure 2). The epithelium was composed of a peripheral layer of palisaded cuboidal cells resting on a basement membrane and several overlying layers of large squamous cells with abundant pale cytoplasm (Figure 3); these cells underwent

abrupt keratinization without the formation of a granular cell layer, giving rise to dense orthokeratotic eosinophilic keratin (Figure 4). Small structures resembling miniature trichilemmal cysts were observed at the base of the tumor. A diagnosis of trichilemmal horn or trichilemmal keratosis was established.

The lesion had been removed completely and thus no further treatment was required.

Cases of Trichilemmal Horn Described in the Literature

	No. of Cases	Sex	Age, y	Site	Clinical Characteristics	Histological Characteristics
Brownstein ³	19	11 women 6 men 2?	6-72 1	9 on the limbs 4 on the back 2 on the face 3 on the scalp 1?		
Kimura ⁶	3	3 men	74, 43, and 55	2 in the right temporal region 1 on the right side of the neck		Electron microscopy: presence of intranuclear inclusion bodies suggestive of HPV
Peteiro ⁸	1	Woman	75	Left preauricular region		Electron microscopy: no intranuclear inclusion bodies
Poblet ⁹	2	2 Women	70 and 78	1 in the right parietal region of the scalp 1 in the right temporal region	Personal or family history of trichilemmal cysts Horn arising from a richilemmal cyst after t spontaneous or surgical drainage	Immunohistochemistry: – Pan-cytokeratin AE1/AE3: ++ – CAM 5.2: – – CD 34: + –S-100 and CEA: –
Dominick ¹⁰	1	Woman	22	Left elbow		
Kudo et al ⁷	1	Woman	53	Left hand	Lesion arising on a burn scar	Electron microscopy: presence of intranuclear inclusion bodies Immunohistochemistry negative for HPV
Michal ⁵	4	4 women	55, 78, 63, and 83	4 on the scalp	Gigantic cutaneous horns	
Nakamura ⁴	2	2 men	70 and 62	Palm of left hand Thumb	Lesions located on skin without hair follicles	
Haro	1	Woman	82	Scalp	Personal history of trichilemmal cysts on the scalp Horn arising on a surgically drained trichilemmal cyst	
Total	34	21 women 11 men 2 ?	Age range: 15 between 13 16 and 5 83 years 1 ?	15 on the head 13 on the extremities 5 on the trunk 1 ?		

Abbreviations: CEA, carcinoembryonic antigen; HPV, human papilloma virus.

Discussion

Trichilemmal horn is an uncommon, benign follicular neoplasm with trichilemmal differentiation. The 2 essential characteristics of the tumor are its clinical presentation in the form of a cutaneous horn and its histology, which shows trichilemmal keratinization at the base of the lesion.

This follicular neoplasm was first described in 1976 by Headington,² who named the lesion trichilemmal keratosis. The term trichilemmal horn was first coined by Brownstein³

in 1979 in a study that described 19 cases of this tumor and defined its essential characteristics. Since that time, 34 cases have been published in the literature, including the one published here (Table).³⁻¹⁰

It presents clinically as an exophytic keratotic lesion. While it can appear on any part of the body, the most common site is the head—15 cases at this site have been published, 12 of them on the scalp and 3 on the face. This is followed by lesions on the limbs, with 13 cases reported, 9 of them by Brownstein, making the limbs the most

common site in his series. Lesions on the palms of the hands have also been reported.⁴

Trichilemmal horns can develop at any age—the age range of published cases is from 16 to 83 years—although they are most common in patients over the age of 50 years. Approximately two-thirds of the cases described have been in women.

The size of the lesions varies, and is usually between 1 cm and 2 cm in diameter. Michal et al,⁵ however, found 4 cases of gigantic trichilemmal horns that reached lengths of up to 25 cm.

The histological characteristics of this tumor are those described above: a squamous cell epithelium composed of a row of palisaded cuboidal cells with the remaining layers composed of large squamous keratinocytes with abundant glycogen in the cytoplasm that causes them to take on a pale color. This epithelium keratinizes abruptly, without forming a granular cell layer, and gives rise to a dense orthokeratotic eosinophilic keratin. This type of keratinization is known as trichilemmal keratinization.

There has been some discussion about the possibility that human papilloma virus (HPV) may be involved in the pathogenesis of this tumor because intranuclear inclusion bodies morphologically similar to HPV have been found in several electron microscopy studies of the neoplasm, such as those carried out by Kimura.⁶ The results of immunohistochemical staining with anti-HPV antibodies, however, have invariably been negative⁷ and such intranuclear inclusion bodies have not been found consistently.⁸

Poblet et al⁹ described 2 cases in which, as in ours, the lesion developed after an attempt to treat a preexisting trichilemmal cyst. Also as in ours, the patients had a personal or family history of multiple trichilemmal cysts. The authors concluded that in some cases this tumor may be caused by the rupture and subsequent marsupialization

of the wall of a trichilemmal cyst. Rather than disappearing, as it does in other cases, the epithelium continues to proliferate, resulting in an exophytic lesion with the morphology of a cutaneous horn. We believe that this may have been the origin of the tumor in the case we describe.

Conflicts of Interest

The authors declare no conflicts of interest.

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