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Dermatology

A Case of a Clinically Proliferating Trichilemmal Cyst Following the Take of Febuxostat and Calcium Carbonate

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Abstract **Case Report**

Trichilemmal cysts or pilar cysts are keratin-filled cysts that originate in the outer root sheath. They are typically found on the scalp of middle-aged women. They are inherited as an autosomal dominant trait. Trichilemmal cysts range from simple trichilemmal cysts to benign and malignant proliferative trichilemmal cysts. Proliferation can be clinical or histological. Clinically, it may present as an increase in size with the onset of an ulceration. Histologically, it translates as the acquisition of certain features which are the dermal proliferation of a squamous epithelium that arranges in a lobular fashion with clear cells containing glycogen in some areas surrounded by a somewhat cellular and glassy stroma; and especially the trichilemmal keratinization in the centre of lobules. There are also certain histopathological criteria to distinguish benign from malignant proliferating trichilemmal cysts. A malignant trichilemmal cyst is characterized by a high mitotic index, atypical mitoses, significant nuclear polymorphism, and tumor invasion of adjacent tissues. Immunohistochemistry can also confirm the benign character by expression of cytokeratin 10 and involucrin. We report here a case of a clinically proliferating trichilemmal cyst following the introduction of calcium carbonate and febuxostat in a patient undergoing treatment for hypertension and renal failure, but retaining a typically simple and benign character on histology.

Keywords: Proliferative trichilemmal cyst – Clinical proliferation – Non proliferative histopathology – Febuxostat – Calcium carbonate.

INTRODUCTION

There is a nosologic difficulty in the classification of trichilemmal cysts. There are simple proliferating benign trichilemmal cysts and proliferating malignant trichilemmal cysts. This classification is based on histologic criteria, although trichilemmal cysts that are histologically typical may sometimes assume a proliferative clinical character. A simple trichilemmal cyst may evolve into a proliferative trichilemmal cyst after trauma or inflammation. We report here the case of a histologically typical trichilemmal cyst that became proliferative after the introduction of febuxostat and calcium carbonate.

CASE REPORT

This 51-year-old patient was treated for arterial hypertension on ramipril and renal failure. She had undergone arteriovenous fistula surgery 02 years ago, and has since been on Kardegic 75mg.

She also developed hypocalcemia and hyperuricemia 06 months ago, for which she was prescribed calcium carbonate 1000mg daily and febuxostat 40mg.

This patient presented with a small epidermoid cyst that had been evolving for 20 years, but since the introduction of calcium carbonate and febuxostat, an increase in cyst size was noted, with the emergence of a horn through the cyst wall (Figure 1) that detached during the shower.

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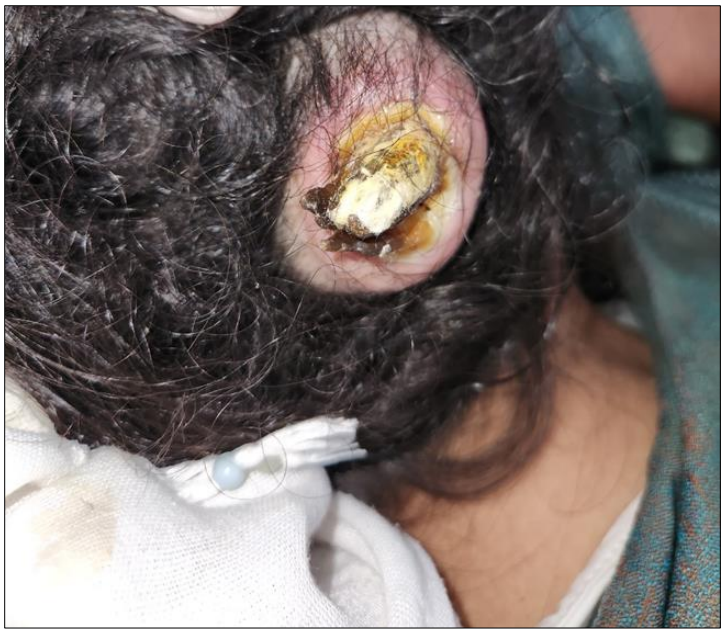

Figure 1: Clinical picture showing a 4cm/3cm smooth-surfaced, non-adherent, painless, soft tumefaction on the occipital scalp, centred by an ulceration crossed by a keratotic horn

On physical examination, the patient presented with a 4cm/3cm smooth-surfaced, non-adherent, painless, soft tumefaction on the occipital scalp, centred by an ulceration filled with sebum and fetid keratotic material (Figure 2).

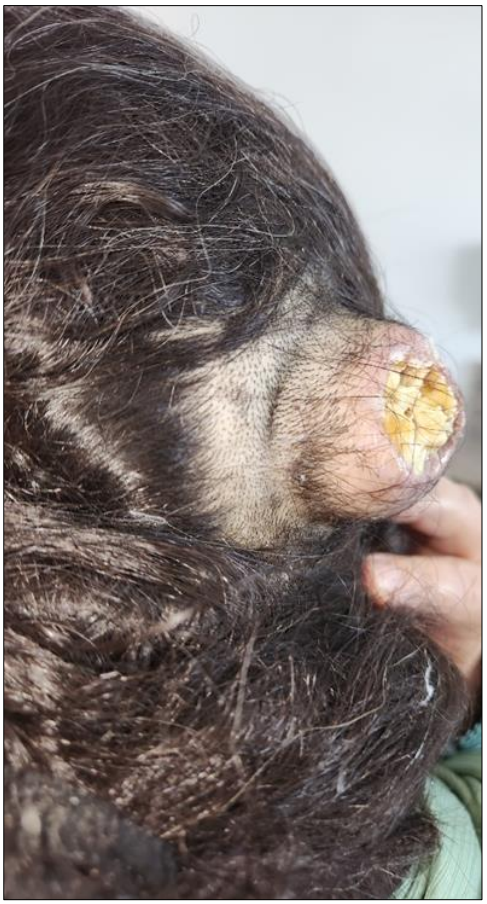

Figure 2: Clinical picture showing a 4cm/3cm smooth-surfaced, non-adherent, painless, soft tumefaction on the occipital scalp, centred by an ulceration filled with sebum and fetid keratotic material

Palpation of the cervical lymph nodes revealed no adenopathy. Dermoscopy of the peri-ulcerous skin showed a patchy brown pigmentation and whitish-pink areas with no vessels (Figure 3).

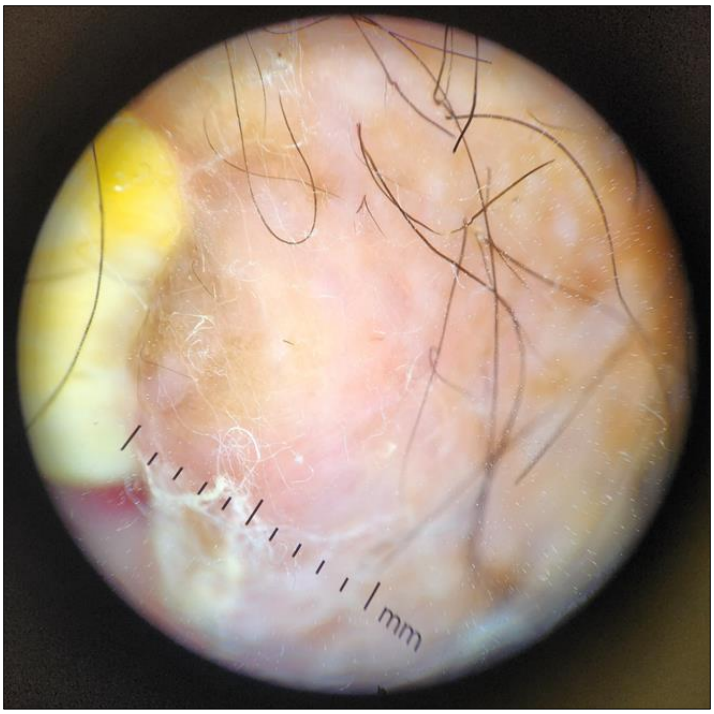

Figure 3 : Dermoscopy of the peri-ulcerous skin showing a patchy brown pigmentation and whitish-pink areas with no vessels (Dermlite DL4 Polarized mode)

Complete excision with pathologic examination revealed a cystic structure bordered by a stratified squamous epithelium with an inner border of globoid cells with abundant acidophilic cytoplasm. The granular layer was absent and the cyst contained dense eosinophilic parakeratotic keratin. In addition, a foreign body-like reaction with giant cells was observed in response to cyst rupture. Calcifications were not noted. All in favor of a trichilemmal cyst with a foreign body-like reaction (Figure 4, 5).

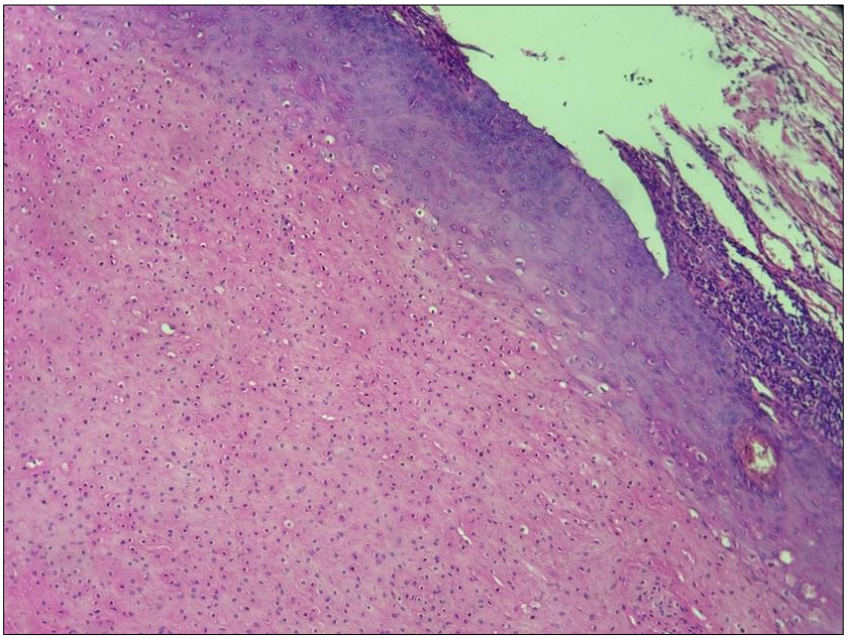

Figure 4 : Histopathology revealing a cystic structure bordered by a stratified squamous epithelium with an inner border of globoid cells with abundant acidophilic cytoplasm. The granular layer is absent and the cyst contains dense eosinophilic parakeratotic keratin (Hematoxylin-Eosin under high magnification)

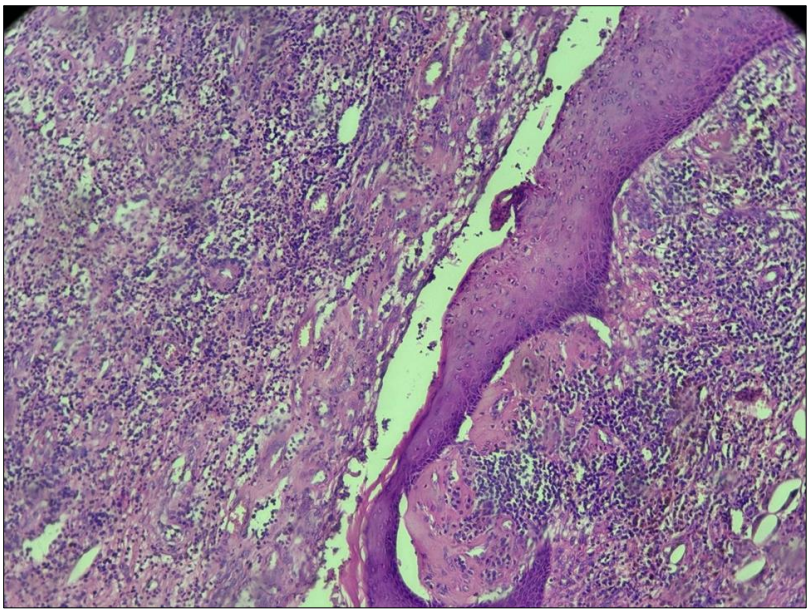

Figure 5 : Histopathology of the cyst showing a foreign body-like reaction with giant cells (Hematoxylin-Eosin under high magnification)

DISCUSSION

Proliferative trichilemmal cyst is a morphologically distinct tumor occurring mainly on the scalp of elderly women. It may be benign or malignant and presents as well circumscribed subepidermal lesions with a uniform histopathological pattern with varying degrees of cytologic atypia.

Proliferative trichilemmal cyst occurs on a common trichilemmal cyst after trauma or inflammation. Ulceration may occur on the trichilemmal cyst. This lesion can be up to 25 cm wide.(Sethi *et al*., 2002)

Histologically, the proliferating trichilemmal cyst or proliferating trichilemmal tumor shows a dermal proliferation of a squamous epithelium that arranges in a lobular fashion with clear glycogen-containing cells in places surrounded by a moderately cellular and glassy stroma.

The main histologic feature is the presence of trichilemmal keratinization in the center of the lobules, defined as the abrupt transition between nucleated and nonnucleated epithelial cells, with the absence of a granular layer.(Noto, 1999; Pereira *et al*., 2023).

The distinction between benign and malignant proliferating trichilemmal cysts is not made clinically but histologically; malignancy is manifested by ill-defined borders, high-grade atypia, aneuploidy, necrosis, cellular pleomorphism, and atypical mitoses(Laing *et al*., 1991; Pereira *et al*., 2023). Both benign and malignant lesions are treated surgically with wide excision margins but there is a risk of recurrence that can be reduced with Mohs micrographic surgery.

Some authors criticize this notion of benign proliferating trichilemmal cyst, arguing that there are proliferating trichilemmal cysts without cytologic atypia that have an aggressive character and vice versa, and that some studies show a histopathological uniformity between so-called benign and malignant trichilemmal cysts(Noto, 1999).

Other authors admit that there is a nosologic difficulty with the concept of benign proliferating trichilemmal cyst, placing it as a form of transition between trichilemmal cyst and malignant proliferating trichilemmal cyst; and show the value of immunohistochemistry, which classifies the tumor as benign when cytokeratin 10 and involucrin are expressed.

Histopathology of a trichilemmal cyst shows a well-defined, subcutaneous or dermal simple cyst, lined by stratified squamous epithelium with a palisading outer layer, containing dense laminated eosinophilic keratin. The granular layer is absent. Calcification is observed in up to 25% of cases. A granulomatous reaction may occur due to cyst rupture. Sebaceous or apocrine glands may be present and the cyst lining resembles that of a hidrocystoma (Ramaswamy *et al*., 2013).

Our patient presented with clinical proliferation but no histopathological evidence of proliferation; thus, she presented with a typical trichilemmal cyst histopathology with foreign body reaction related to the spillage of sebaceous and keratotic material within the scalp stroma. This particular case highlights the possibility of a purely clinical proliferation and supports the idea that the proliferating trichilemmal cyst is a transitional form between the simple trichilemmal cyst and the malignant proliferating trichilemmal cyst.

Treatment is surgical and, in the case of malignant proliferative trichilemmal cysts, may be combined with cisplatin and 5-FU-based chemotherapy followed by palliative radiotherapy under close observation due to the high risk of recurrence and metastasis (Kamal *et al*., 2021).

CONCLUSION

Our patient presented with a trichilemmal cyst of increasing size, simulating a proliferative trichilemmal cyst arising outside the context of trauma or inflammation. Our case is the first to describe the clinical proliferative character in relation to the introduction of a drug, specifically febuxostat and calcium carbonate. The pathological anatomy showed no evidence of malignancy; however, benignity could have been confirmed by immunohistochemistry showing expression of cytokeratin 10 and involucrin.

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REFERENCES

- Kamal, I. B., Hali, F., Marnissi, F., & Chiheb, S. (2021). The Proliferating and Malignant Proliferating Trichilemmal Cyst: An Anatomo-Clinical Study of Three Cases. *Skin Appendage Disorders*, *8*(2), 161. https://doi.org/10.1159/000518354
- Laing, V., Knipe, R. C., Flowers, F. P., Stoer, C. B., & Ramos-Caro, F. A. (1991). Proliferating trichilemmal tumor: Report of a case and review of the literature. *The Journal of Dermatologic Surgery and Oncology*, *17*(3), 295-298. https://doi.org/10.1111/j.1524-4725.1991.tb03646.x
- Noto. (1999). 'Benign' proliferating trichilemmal tumour : Does it really exist? *Histopathology*, *35*(4), 386-387. https://doi.org/10.1046/j.1365-2559.1999.00747.x
- Pereira, A. A., Bueno, J. L., Boff, A. L., & Souza, P. R. M. (2023). Proliferating trichilemmal tumor. *Anais Brasileiros de Dermatologia*, *98*(6), 864. https://doi.org/10.1016/j.abd.2021.11.015
- Ramaswamy, A. S., Manjunatha, H. K., Sunilkumar, B., & Arunkumar, S. P. (2013). Morphological Spectrum of Pilar Cysts. *North American Journal of Medical Sciences*, *5*(2), 124. https://doi.org/10.4103/1947-2714.107532
- Sethi, S., & Singh, U. R. (2002). Proliferating trichilemmal cyst: Report of two cases, one benign and the other malignant. *The Journal of Dermatology*, *29*(4), 214-220. https://doi.org/10.1111/j.1346-8138.2002.tb00252.x