

Giant Solitary Trichoepithelioma: A Diagnosis of Exclusion

Sir,

Trichoepitheliomas (TEs) are regarded as poorly differentiated hamartomas of hair germ.^[1] There are three variants of TE, namely solitary, multiple, and desmoplastic. When a solitary TE is 2 cm or more in size, then it is labeled as giant solitary trichoepithelioma (GST).^[2]

GST is quite an uncommon appendageal tumor to permit a ready familiarity. We hereby report a case of GST, whose diagnosis was arrived primarily by the histopathological study as a diagnosis of exclusion, because both clinical and dermoscopic findings were not very conclusive for a single entity.

A 65-year-old female presented to the dermatology outpatient department with complaints of a slowly expanding asymptomatic lesion over the forehead for the past 10 months. It began as a minuscule pea-sized papule that slowly advanced to attain the current status [Figure 1a]. On examination, an indurated, annular plaque with asymmetric thickened borders was visualized. The border was broadened over the right margin with the thickening going on to enclose almost 80% of the plaque leaving behind a central depression of size 0.3 cm × 0.3 cm. Toward the upper and outer right border of the plaque, overlying crusting/scaling was seen. The left border was considerably thinner when compared to the right side [Figure 1b]. Palpation demonstrated a



Figure 1: (a) An annular sclerotic plaque on the forehead as seen on presentation (b) A closer view of the plaque demonstrating an asymmetrical thickening of the border with a crusted area on the right upper and outer aspect and a central depression

firm and nontender consistency. Clinical differentials of desmoplastic TE and sclerotic basal cell carcinoma (BCC) were considered. Dermoscopy revealed short, thick white crossing lines (consistent with chrysalis pattern), few telangiectatic vessels, circular white to cream-colored structures [Figure 2a] and yellowish-brown crusts and white scales along with arborizing vessels [Figure 2b].

Skin biopsy demonstrated an atrophic epidermis. Dermis was composed of islands of uniform basilar cells with peripheral palisading in few areas, and numerous keratinous cysts lined by stratified squamous epithelium. The basophilic cell nests lacked retraction clefts. Many of these nests demonstrated abortive hair follicle differentiation. The stroma exhibited a fibromyxoid consistency [Figures 3 and 4]. As facilities for cytokeratin markers were not available in our institute, this test could not be performed. Nevertheless, on careful evaluation of clinical, dermoscopic, and histopathological findings, a diagnosis of GST was finally arrived at.

Although the clinical morphology in our case was highly consistent with desmoplastic TE (illustrating the characteristic annular, indurated, and centrally depressed plaque); histopathology failed to demonstrate two components of the classical histopathological triad diagnostic for desmoplastic TE (viz. narrow strands of basaloid tumor cells and a desmoplastic stroma) as stated by Brownstein and Shapiro.^[3] Only one component of the triad, i.e., keratinous cysts was identified, thereby excluding its diagnosis.

Histopathology, in our case, elaborated a high degree of differentiation toward follicular structures along with

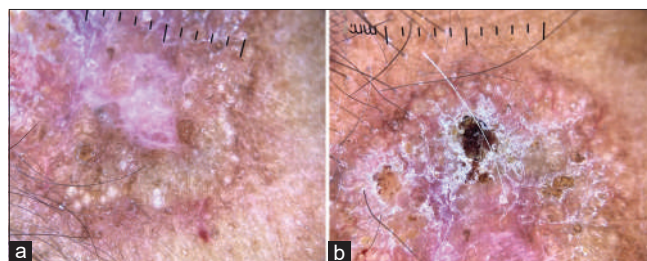


Figure 2: (a) Dermoscopic evaluation showing a chrysalis pattern, along with telangiectatic vessels and circular white to cream-colored structures (b) Dermoscopic evaluation showing yellowish-brown crusting and white scaling along with arborizing vessels

1 numerous horn cysts and abortive dermal papillae. Also, the
 2 absence of mitotic figures and a noninfiltrative pattern on
 3 microscopy further pointed toward a diagnosis of solitary
 4 TE.^[4] The only finding causing doubt was the unusual
 5 clinical manifestation of the adnexal tumor. This sclerotic
 6 presentation of GST, we believe has not been previously
 7 reported and we therefore would like to highlight this
 8 new morphological pattern that we observed. Other rare
 9 phenotypes of GST that have been described in the past
 10 include subcutaneous nodules,^[5] pedunculated plaques,^[6]
 11 ulcerated forms,^[7] and cystic lesions.^[8]

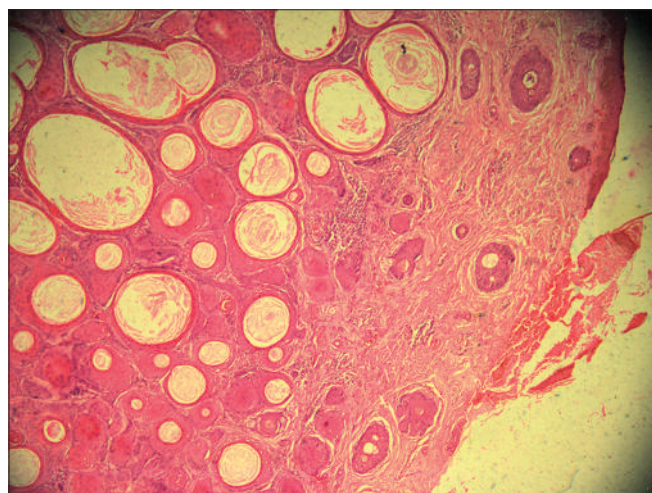


Figure 3: Histopathology demonstrating epidermal atrophy with aggregates of basaloid cells delineating abortive hair follicle differentiation to a high degree. One area illustrates early follicular stromal induction within nests of basaloid cells. Nests of basaloid cells lack retraction clefts. Furthermore, numerous horn cysts lined by stratified squamous epithelium can be observed (H and E, $\times 10$)

Besides, we discovered that dermoscopy was not a very specific diagnostic tool here. Nevertheless, it enabled studying the underlying dermal pathology better. A number of findings overlapping with BCC/other skin adnexal tumors were identified in our dermoscopic evaluation and have been elaborated in Table 1, which we consider to be of diagnostic value while evaluating adnexal tumors. However, histopathology still remains the gold standard

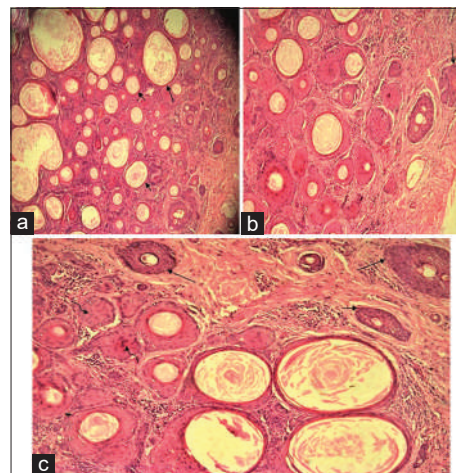


Figure 4: (a) Section demonstrates numerous horn cysts (straight black arrows) along with aggregates of basaloid cells (H and E, $\times 10$) (b) Section illustrates aggregates of basaloid cells and the presence of a focus showing early follicular stromal induction within nests of aggregated basaloid cells (straight black arrow) (H and E, $\times 10$) (c) Section elucidates numerous horn cysts surrounding which is a moderately dense lymphocytic infiltrate. A fibromyxoid stroma can be observed. Furthermore, aggregates of basaloid cells with peripheral palisading can be seen (curved black arrows) and a high degree of differentiation of basaloid cells toward follicular structures can also be observed (straight black arrows) (H and E, $\times 20$)

Table 1: Analysis of dermoscopic patterns as seen in our case and in other conditions where such patterns could be encountered

Pattern	Description	Cause	Other conditions where seen	Remarks
Chrysalis pattern ^[9]	Short, thick, bright, white orthogonally oriented linear streaks	Underlying dermal fibrosis	Melanoma Spitz nevi BCC Dermatofibroma Scars Desmoplastic TE	Has not been previously reported in GST This could be attributed to the indurated character of the lesion; although the amount of sclerosis within the dermis was not very significant in our patient More dermoscopic evaluations need to be done, to strengthen our observation
Arborizing vessels ^[10]	Sharp, bright red large stem vessels branching into fine terminal capillaries	Dilated neovasculature in the superficial dermis	BCC Desmoplastic TE TE	Was identified in our case
Fine telangiectasia ^[10]	Short, fine, linear blood vessels	Telangiectatic vessels in the superficial dermis	BCC Desmoplastic TE Microcystic adnexal carcinoma TE	Was identified in our case
Circular white to cream-colored structures ^[11]	Opaque to white, round to oval circumscribed areas	They represent keratin cysts and calcification within the tumor	Desmoplastic TE Microcystic adnexal carcinoma TE	Was identified in our case
Yellow-brown crusts ^[10]	Well defined brownish yellow to white scales	Thin crusts overlying superficial epidermal loss	BCC	Could be a nonspecific finding in our case

TE – Trichoepithelioma; GST – Giant solitary trichoepithelioma; BCC – Basal cell carcinoma

for diagnosing these appendageal tumors, the strength of which can be enhanced by immunohistochemistry.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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REFERENCES

- Anderson DE, Howell JB. Epithelioma adenoides cysticum: Genetic update. *Br J Dermatol* 1976;95:225-32.

- Teli B, Thrishuli PB, Santhosh R, Amar DN, Rajpurohit S. Giant solitary trichoepithelioma. *South Asian J Cancer* 2015;4:41-4.
- Brownstein MH, Shapiro L. Desmoplastic trichoepithelioma. *Cancer* 1977;40:2979-86.
- Zeligman I. Solitary trichoepithelioma. *Arch Dermatol* 1960;82:35-40.
- Swaroop MR, Sathyanarayana BD, Chaurasia PR, Devaraj Y, Dukkupati M, Sajid A. Giant solitary trichoepithelioma over the nose. *Indian J Dermatopathol Diagn Dermatol* 2017;4:17-8.
- Krishnamurthy J, Divya K. The cytology of giant solitary trichoepithelioma. *J Cytol* 2010;27:99-101.
- Goyal S, Mahajan NC, Garg M, Goyal S. Giant solitary nodular trichoepithelioma. A case report and review of literature. *Arch Clin Exp Surg* 2012;1:58-60.
- Lorenzo MJ, Yebera-Pimentel MT, Peteiro C, Toribio J. Cystic giant solitary trichoepithelioma. *Am J Dermatopathol* 1992;14:155-60.
- Balagula Y, Braun RP, Rabinovitz HS, Dusza SW, Scope A, Liebman TN, *et al.* The significance of crystalline/chrysalis structures in the diagnosis of melanocytic and nonmelanocytic lesions. *J Am Acad Dermatol* 2012;67:194.e1-8.
- Cameron MC, Lee E, Hibler BP, Giordano CN, Barker CA, Mori S, *et al.* Basal cell carcinoma: Contemporary approaches to diagnosis, treatment, and prevention. *J Am Acad Dermatol* 2019;80:321-39.
- Costello CM, Han MY, Severson KJ, Maly CJ, Yonan Y, Nelson SA, *et al.* Dermoscopic characteristics of microcystic adnexal carcinoma, desmoplastic trichoepithelioma, and morpheaform basal cell carcinoma. *Int J Dermatol* 2021;60:e83-4.

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Letter to Editor

Serum Paroxonase 1 Level and Androgenetic Alopecia: Correspondence

Sir,

We would like to share ideas on “Serum paroxonase 1 level may be an indicator and predictor of the severity of androgenetic alopecia.^[1]” Tantawy *et al.* concluded that “the level of passive optical network 1 significantly decreased in AGA patients, which may give additional proof that OS has role in the pathogenesis of AGA and hence may help in the management of AGA by adding antioxidants in the treatment.^[1]” We agree that serum paroxonase 1 level might be a useful biomarker. However, serum paroxonase 1 level is affected by the many factors. Alteration of serum paroxonase 1 is observed in cases with diabetes and dyslipidemia.^[2] In addition, genetic polymorphism background also affects serum paroxonase 1 level.^[3] If Tantawy *et al.* plan for additional further study, the assessment of the mentioned confounding factors is interesting.

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Authors ask for waiving for any charge for this correspondence.

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REFERENCES

1. Tantawy M, Khabir AA, Mahsoub N, Zohdy M. Serum paroxonase 1 level may be an indicator and predictor of the severity of androgenetic alopecia. *Int J Trichology* 2021;13:26-31.
2. Dullaart RP, Otvos JD, James RW. Serum paroxonase-1 activity is more closely related to HDL particle concentration and large HDL particles than to HDL cholesterol in Type 2 diabetic and non-diabetic subjects. *Clin Biochem* 2014;47:1022-7.
3. Kulka M. A review of paroxonase 1 properties and diagnostic applications. *Pol J Vet Sci* 2016;19:225-32.

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