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Case Report

Pathology Section

A Large Recurrent Proliferating Trichilemmal Tumour of Gluteal Region: A Case Report with Review of Literature

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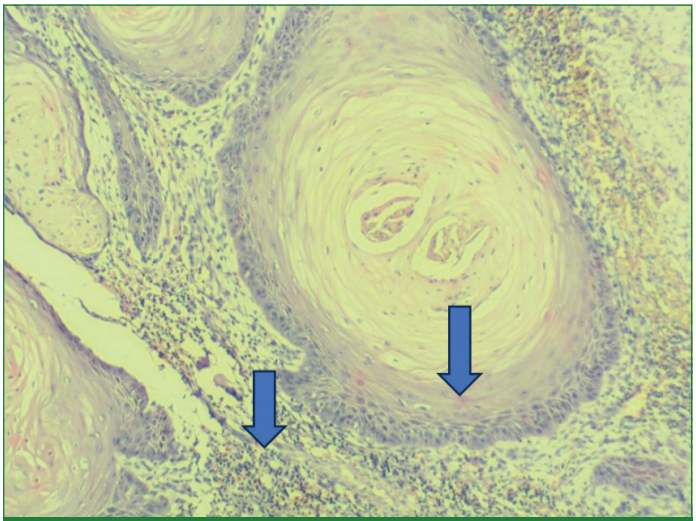
ABSTRACT

Proliferating Trichilemmal Tumour (PTT) is an uncommon, benign skin appendageal tumour with distinctive morphologic features. It most commonly presents as a well-circumscribed subcutaneous nodule, and its histological hallmark is the presence of trichilemmal keratinisation and squamoid cytological features. They are believed to originated from the Trichilemmal Cyst (TC) and have malignant potential, termed a Malignant PTT (MPTT). These lesions may cause significant morbidity and have a risk of recurrence after simple local excision. The lesion mimics morphologic resemblance to squamous cell carcinoma at the microscopic level, making a correct histologic diagnosis extremely important. The present case involves a large PTT occurring in the gluteal region of a 67-year-old man, which is an uncommon anatomical location. The tumour recurred despite previous excision, indicating its potential for local aggressiveness. The large size of lesion, which clinically mimicked malignancy. Despite clinical suspicion, histopathology did not meet malignant criteria, helping distinguish it from squamous cell carcinoma or MPTT. This underscores the importance of correct histopathological interpretation, especially when diagnostic challenges arise due to morphological resemblance to squamous cell carcinoma.

Keywords: Benign skin adnexal tumour, Malignant transformation, Squamoid cytological features, Subcutaneous nodules, Trichilemmal keratinisation

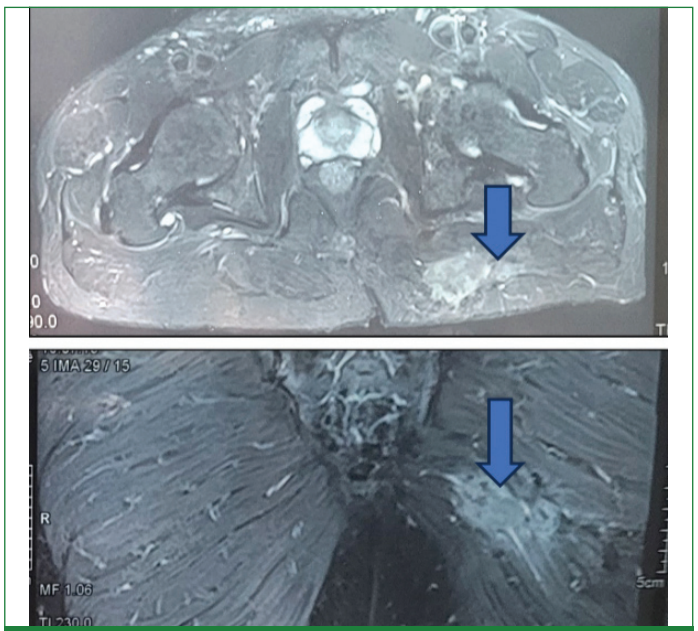
CASE REPORT

A 67-year-old man with a painless recurrent gluteal mass on the left-side for two years. He had a history of recurrent left gluteal mass operated thrice over eight years, histologically reported as an epidermal inclusion cyst. There were not significant medical comorbidities noted. On examination healthy man without lymphadenopathy except recurrent swelling at the same site after a long span which was excised and histologically diagnosed as a PTT [Table/Fig-1]. Again, he developed recurrent swelling at the same site five months after the second time operation.



[Table/Fig-1]: Haematoxylin and Eosin (H&E) 100x- Trucut biopsysection shows cyst wall lined by squamous epithelial lining with trichilemmal keratinisation (indicated by arrow) & dense lymphocytic response around the nests of squamous epithelial cells.

MRI of the pelvis [Table/Fig-2] showed ill-marginated soft-tissue organised collection noted in the left-side's gluteal musculature and subcutaneous area. It measures approximately 4.5×2.4×3.4 cm. The collection appears heterogeneously hyperintense on T2-weighted images. On postcontrast scan, serpiginous enhancement of thin wall noted. On the Diffusion-Weighted Imaging (DWI) sequence, the content shows diffusion restriction. The overlying gluteal fat showed inflamed changes, and the visualised soft-tissues were normal in morphology and signal intensity. No areas of calcification or solid–cystic components were identified, making a trichilemmal tumour unlikely. The imaging findings were suggestive of an abscess or haematoma within the left gluteal musculature.

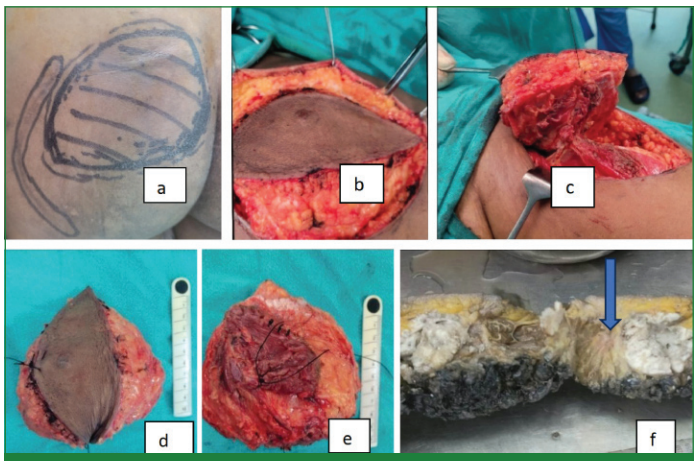


[Table/Fig-2]: MRI Pelvis shows: 1) upper image- ill-marginated soft-tissue organised collection noted in the left-side's gluteal musculature and subcutaneous area Indicated by blue arrow; 2) lower image shows soft-tissue mass infiltrating the gluteal musculature (post-contrast).

A trucut biopsy was done from the left gluteal mass, obtaining eight friable fragmented tissue cores, the largest measuring 0.4 cm in aggregate measuring 0.4×0.4×0.1 cm (TS). The section showed fragmented cores with acanthotic hyperkeratotic squamous

epithelium, along with cores showing benign fibrostromal connective tissues. Features were suggestive of a trichilemmal tumour, with no evidence of malignant transformation. The case was discussed in the tumour board because of recurrent proliferating trichilemmal tumour at the same site for a consensus opinion. The board decision was given to do frozen biopsy for wide local excision with advised to know the status surgical margin ether positive or negative for tumour.

Perioperative findings [Table/Fig-3] were an irregular nodule measuring 10×6 cm in the left buttock, fixed to underlying muscles and part of the overlying skin, followed by wide local excision with adequate soft-tissue base margin and skeletal muscle.



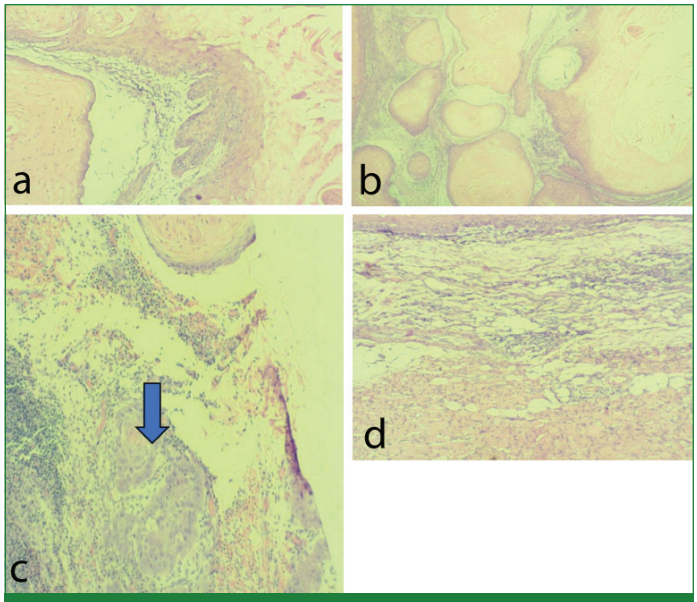
[Table/Fig-3]: a) Shows skin markings during perioperative period; b) Intraoperatively dissection of lesion; c) excision of lesion with skeletal muscle; d) wide local excision specimen; e) wide local excision with base margin; f) Gross specimen shows an irregular nodule measuring 10×6 cm in the left buttock, fixed to underlying muscles and part of the overlying skin. (Arrow indicates -tumour part).

The specimen was sent for a frozen section; found that all margins are free of tumour more than 1 cm away from lesion.

The gross specimen showed a skin-covered nodular mass measuring 11×9×5.5 cm, overlying elliptical skin measuring 10×7×0.2 cm with suture-tied margins. On cut section, a well-circumscribed mass measuring 4.5×4×4 cm was identified. The lesion was predominantly cystic, containing abundant cheesy white pultaceous material. Approximately one-third of the tumour was solid, appearing whitish to cream in colour, with focal areas of cystic change.

The distances of the tumour from the resection margins are as follows: base margin- 1 cm; lateral margin- 2 cm; medial margin- 2.5 cm; superior soft-tissue margin- 3.2 cm; inferior soft-tissue margin- 1.8 cm; superior skin margin- 1.5 cm; inferior skin resected margin- 1.2 cm. The overlying skin is 1 cm away from the tumour.

Stained H&E sections showed proliferation of squamous epithelial cells forming nests with trichilemmal keratinisation and chronic inflammatory response in stroma. At focal areas, stromal invasion by atypical squamous cells was identified. The histomorphological features are suggestive of a proliferating trichilemmal tumour (PTT) with foci of minimally invasive squamous cell carcinoma [Table/Fig-4]. All resection margins are free of tumour. On follow-up, the patient is doing well and has received six sessions of radiotherapy to reduce the risk of recurrence at the time of this report.



[Table/Fig-4]: H & E X100- Excisional biopsy of gluteal mass & its section shows: a) Hyperplastic squamous epithelial lining with mostly intact basement membrane & laminated keratin; b) Irregular nests of proliferated squamous epithelium; c) Invasive nests of squamous epithelium with atypia & peritumoural inflammatory response; d) dense chronic inflammation in subepithelial fibro stromal adipose tissue.

DISCUSSION

The PTT are rare benign skin adnexal tumours, including proliferating pilar tumours, proliferating trichilemmal cysts, or proliferating follicular-cystic neoplasm with follicular isthmic differentiation and varying degrees of atypia. Malignant transformation is a rarer pathological finding and most of these tumours arise within the wall of a pre-existing trichilemmal cyst. These tumours are most commonly seen in sun-exposed areas and regions with abundant hair growth, which explains their highest frequency on the scalp [1]. They are less commonly reported on the trunk, forehead, nose, back, chest, abdomen, buttocks, elbow, wrist, mons pubis, and vulva [2].

The PTTs are mostly found in women, 80-87% of the cases, between 27 and 83 years of age, with a peak in the sixth and seventh decades of life [3,4]. Moreover, only two cases in the literature have reported the presence of an MPTT over the skin of the breast [5,6].

Histologically, these are characterised by an abrupt transition of the nucleated epithelium to a nucleate keratinised cells without a granular layer, known as trichilemmal keratinisation. In most cases, MPTTs can occur due to either de novo or malignant transformation of a PTT. A stepwise transformation of MPTTs has been described from an adenomatous to an epitheliomatous and then to a carcinomatous stage [7].

Recent literature suggests that recurrent lesions [Table/Fig-5] share several common features: they most often arise on the scalp of older patients, present as long-standing nodules with recent rapid growth, and tend to recur following prior limited or incomplete excision [8-12]. In contrast, malignant and atypical cases demonstrate deeper invasion, higher rates of regional and distant metastasis, and more frequent use of radiotherapy or systemic therapy. In the present case, the lesion represented a recurrent proliferating trichilemmal tumour (PTT) with foci of minimally invasive squamous cell carcinoma, which

S. no	Author and publication year	Age/Gender	Site of lesion	Trucut biopsy	Excisional biopsy	Recurrence
1	Adegun OK et al., 2019 [8]	70 years/F	Left lateral nape of neck	SCC	PTT	Not present
2	Miyachi H et al., 2016 [9]	94 years/F	Scalp	nil	PTT	Not present
3	Fonseca TC et al., 2016 [10]	80 years/F	Scalp	nil	PTT	Present
4	Mohan B et al., 2020 [11]	85 years/F	Scalp	nil	MPTT	Not present
5	Capurso-Garcia MA et al., 2016 [12]	39 years/F	Skin over breast	nil	PTT	Present
6	Present case, 2026	67 years/M	Gluteal region	Trichilemmal tumour	PTT with minimal infiltrating SCC	Present

[Table/Fig-5]: Reported cases of Proliferating Trichilemmal Tumour (PTT) and comparison with present case [8-12].
SCC: Squamous cell carcinoma; PTT: Proliferating trichilemmal tumour; MPTT: Malignant proliferating trichilemmal tumour

tend to remain locally confined and can be effectively managed with wide local excision and adjuvant radiotherapy.

A study by Alici O et al., in which 76 patients were evaluated, proposed a clinicopathological categorisation of proliferating trichilemmal tumours (PTTs) into three groups [13]:

(i) Group 1: Benign tumours showing minimal nuclear atypia, trichilemmal keratinisation, and stromal invasion associated with chronic inflammatory response and multinucleated giant cells.

(ii) Group 2: Locally aggressive tumours characterised by irregular, locally invasive contours, moderate cytological atypia, foci of single-cell necrosis, abrupt keratinisation, and a desmoplastic stroma.

(iii) Group 3: Malignant tumours exhibiting the above features along with marked pleomorphism, atypical mitotic figures, and lymphovascular invasion.

The morphologic resemblance of PTT with well-differentiated squamous cell carcinoma often poses a diagnostic challenge to histopathologists. The tumour cells in an otherwise classical PTT may show focal areas of nuclear atypia and individual cell keratinisation, which at first glance suggest squamous cell carcinoma. Diagnostic difficulties were: 1) the presence of abrupt keratinisation, minimal pleomorphism, low mitotic activity, sharp circumscription, indistinguishable from a trichilemmal cyst with areas of calcification. A trichilemmal cyst was ruled out in view of the presence of epithelial nests and absence of calcification. Premalignant lesions such as actinic keratosis, which may aid in differentiating PTT from squamous cell carcinoma [14,15], were also excluded due to the absence of irregular acanthosis and basal layer atypia; 3) In rare instances, PTT can undergo malignant transformation, indicated by rapid nodule enlargement. Histologically, MPTT s show severe nuclear atypia, marked cellular pleomorphism with atypical mitoses, dyskeratotic cells, and infiltrating margins [16]. A combination of diverse features such as non-scalp location, recent rapid growth, size greater than 5 cm, infiltrative growth, and significant cytologic atypia with mitotic activity favour a diagnosis of malignant PT [17]. In the present case, the lesion measured less than 5 cm and showed no significant cytological atypia or increased mitotic activity, thereby ruling out malignant proliferating trichilemmal tumour (MPTT). Mutations in the p53 gene have been reported in some cases [18], although other studies have not demonstrated p53 protein overexpression. Lymph node and distant metastases have been described in MPTTs. Surgical excision with adequate wide margins remains the treatment of choice for both benign and malignant PTTs.

The PTTs with focal invasion require adequate treatment (wide local excision) and follow-up (long term), particularly in large and long-standing lesions [19]. However, both forms are known to recur even after adequate resection. Chemotherapy and radiotherapy have also been used for malignant tumours [20].

In the present case, recurrence of the lesion was noted in the gluteal region one year after the initial diagnosis of proliferating trichilemmal tumour. It was preceded by a trichilemmal cyst diagnosed multiple times over seven years. Because of the recurrence of the lesion, the possibility of soft-tissue sarcoma might be considered. Fine Needle Aspiration (FNA) gluteal mass shows a low-grade squamous proliferative lesion. The core needle biopsy report for the first time was inconclusive. An MRI of the pelvis showed a deep-seated lesion with an organised collection of ill-defined margins, considered as an abscess/haematoma in gluteal musculature and subcutaneous areas. Because of clinical radiological disparity and inconclusive reports on core needle biopsy leading to diagnostic dilemma. The frozen biopsy report was that all margins were free of tumours. The histology report consists of a predominance of PTTs with significant cytological atypia and subtle minimal stromal invasion. However, entrapped nests of squamous cells mimic stromal invasion. The final diagnosis was foci of minimal squamous cell carcinoma in a background of PTT. Despite this, the histological criteria for diagnosing malignant proliferating trichilemmal tumour (MPTT) were not met in this case.

Immunohistochemistry can be used to distinguish MPTTs from PTTs and SCCs. CD34 is a marker closely associated with trichilemmal keratinisation, so it is positive in MPTT and negative in SCCs. Also, MPTTs have a greater chance of recurring and metastasising [21]. Some authors assume that the loss of CD34 staining is related to a decrease in tumour differentiation. Ki-67 and p53 can help distinguish between MPTTs and PTTs, as both markers are usually absent or dimly expressed in PTTs. IHC was not done in this case due to histomorphology diagnosis of PTT because of few tumour nests (<5%) in the stroma. In view of the minimal invasive component (<5%), the findings were insufficient to fulfil the diagnostic criteria for malignancy. Immunohistochemistry (IHC) was considered unlikely to provide additional diagnostic value or alter clinical management and was therefore deferred. The differential diagnoses included proliferating trichilemmal tumour (PTT) with foci of minimally invasive squamous cell carcinoma and malignant PTT. IHC markers such as CD34, Ki-67, and p53 may aid in further characterisation, if required. Additionally, the similarities in histopathological features of these entities can make evaluation challenging for the reporting pathologist. PTT is presumed to arise from trichilemmal cysts and may progress to a malignant PTT. Both tumours can closely mimic squamous cell carcinoma [22]. Therefore, correlation with histopathology is critical to obtain the best patient outcome and determine if further adjuvant therapy is required. In present case, a wide local excision of the tumour with a 1 cm margin of normal tissue was done. Further radiotherapy was given to prevent the local recurrence and distant metastasis.

CONCLUSION(S)

The PTTs can be presented at uncommon locations and increased morbidity, which constitutes a challenging situation clinically, making histological diagnosis and level of treatment due to the nonspecific clinical presentation and rarity of this condition. Incompletely excised margins may have a chance of lesion recurrence for long periods, transforming to malignancy. A multidisciplinary approach, including tumour board discussion, guided the management decision, following which wide local excision with 1 cm tumour-free margins was performed to reduce the risk of recurrence and distant metastasis.

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