Case Report

Gluteal Malignant Proliferating Pilar Tumor: An Unusual Presentation in an Elderly Male

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Abstract

Malignant proliferating trichilemmal tumor, though uncommon, predominantly occurs in the scalp, trunk, head, and neck, and females are more commonly affected. It is rarely seen in the extremities, and malignant transformation is a rarity. Due to the rarity of malignant transformation, only a hand full of cases has been documented in the literature. We present an unusual case of gluteal malignant proliferating pilar tumor in an elderly male.

Keywords: Gluteal, malignant, pilar, squamous cell carcinoma, trichilemmal

Introduction

Pilar tumor of the scalp (also known as proliferating pilar/trichilemmal cyst) is a rare benign locally aggressive skin adnexal tumor arising from the hair follicle outer root sheath.[1,2] Proliferating trichilemmal cyst, though uncommon, predominantly occurs in the scalp, trunk, head, and neck, and females are more commonly affected.[1-3] It is rarely seen in the extremities, and malignant transformation is a rarity. Due to the rarity of malignant transformation, only a hand full of cases has been documented in literature. We present an unusual case of gluteal malignant proliferating pilar tumor in an elderly male.

Case Report

A 69-year-old well-nourished elderly male presented to a private hospital with a 10-year history of progressive swelling of the left buttock. There was no antecedent history of trauma or surgery to the site of swelling, which became painful a couple of months before presentation at the private hospital. He also complained of rapid increase in the size of the swelling in the past 2 years preceding hospital presentation. There were no other visible or palpable swellings on his body. He had no significant past medical history or constitutional symptoms suggestive of any systemic diseases. Clinical examination of the left buttock showed a circumscribed firm gluteal mass measuring 12 cm × 8 cm and adherent to the underlying structures and inferior margin of the gluteus. The mass was covered by intact skin. There were no palpable masses in the groin or anywhere else. A clinical impression of gluteal lipoma was made, and an excisional biopsy was sent to the pathology laboratory for analysis.

Grossly, we received an irregular lobulated cystic mass with an attached skin; the specimen measured 13 cm × 8 cm × 6 cm and weighed 158 g [Figure 1]. Cut surfaces of the mass revealed a cystic cavity with gray thickened wall containing friable materials. Other areas on the cut surfaces were gray and homogenous [Figure 2].

Microscopic examination showed a keratinized skin overlying a deep dermal cyst lined by keratinized stratified squamous epithelium. The cyst contained lamellated keratin within the cyst cavity. An infiltrative well-differentiated tumor arrayed in nests, trabeculae, and small sheets which arose from the wall of the cyst. It was composed of malignant polygonal cells having hyperchromatic nuclei with 1–2 nucleoli and a moderate amount of amphophilic cytoplasm admixed with clear cells, central keratin formations, multinucleate giant cells, and abnormal mitotic figures [Figures 3 and 4]. The deep resection margins

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are involve by tumour [Figure 5]. A diagnosis of malignant proliferating pilar (trichilemmal) tumor with surgical margin involvement was made.

Discussion

Malignant proliferating pilar (trichilemmal) tumor is a rare malignant skin adnexal tumor of hair follicle origin and was recognized by Saida *et al*. in 1983 as a distinct entity arising from an uncommon benign pilar tumor or proliferating trichilemmal cyst of the scalp.[4] The benign counterpart was first described by Jones[5] in 1966 as proliferating epidermoid tumor and it predominantly affects middle aged to elderly females with a male-female ratio of 6:1 and has a predilection for the scalp.[1-4] However, other sites of involvement, particularly, terminal hair-bearing regions of the body have been documented in literature.[3,6] It often presents as a solitary multinodular deep dermal tumor which may extend to the subcutaneous tissue and has an average size of 5 cm though ranges from 2 to 10 cm. Malignant transformation was highlighted by Headington[7] and is often associated with lesions >6 cm in size, rapid growth, surface ulceration, and locations other than the scalp.[8-10] Our patient is male and presented with a 13 cm gluteal mass with associated pain and rapid growth in the past 2 years before presentation. Although he presented a lump in the buttocks of 10-year duration, it was the sudden rapid increase in size that prompts him to seek medical attention. An impression of lipoma was made by the attending surgeon.

There is no racial, geographic distribution, or known risk factor for the development of malignant proliferating pilar tumor and the incidence is unknown. This uncertain incidence rate may be related to the erroneous misdiagnosis of proliferating pilar tumor as other skin adnexal tumors and skin cancers, particularly, squamous cell carcinoma due to the similarity of some histological features with these other lesions, particularly lesions with trichilemmal type keratinization such as epidermoid cyst, pilomatricoma, trichilemmal pilar tumor and proliferating pilar tumor, hair matrix tumor, and squamous cell carcinoma.[3,11]

Definitive diagnosis is achieved by tissue biopsy and identification of a deep dermal trichilemmal cyst in continuity with an infiltrative malignant tumor exhibiting marked

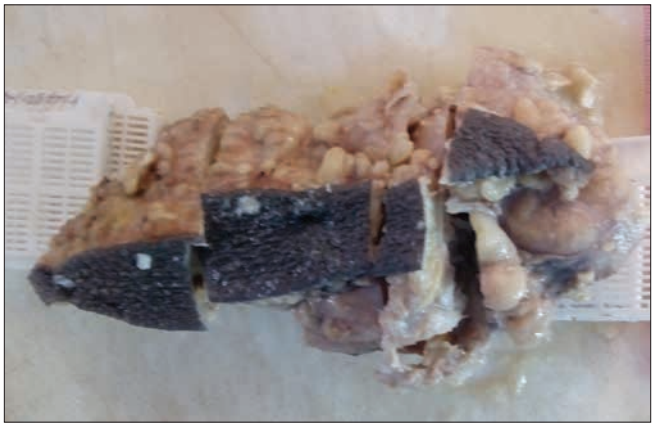

Figure 1: Skin covered biopsy from the gluteal area

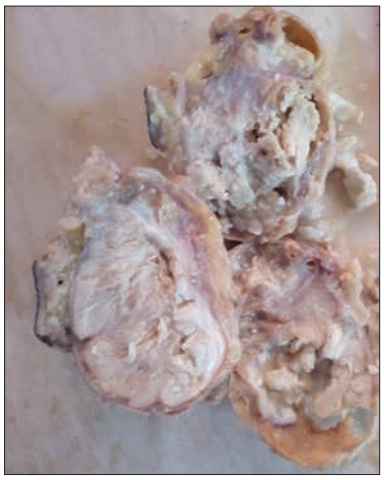

Figure 2: Cut surfaces of the mass revealed a cystic cavity with gray thickened wall containing friable materials

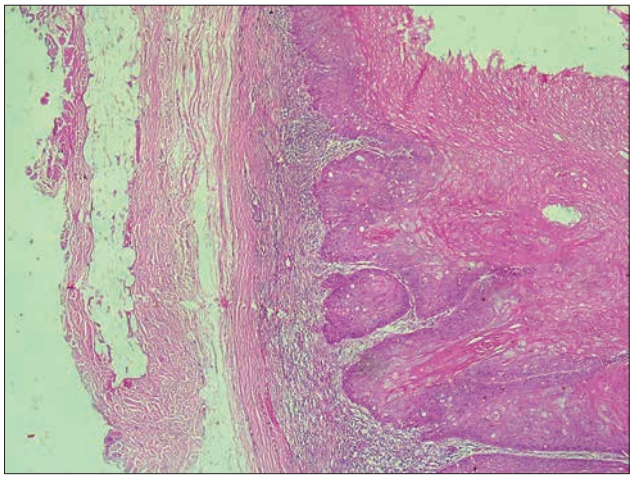

Figure 3: Histological section shows a cystic cavity containing keratin and lined by lobules and nests of well differentiated tumour, ×200

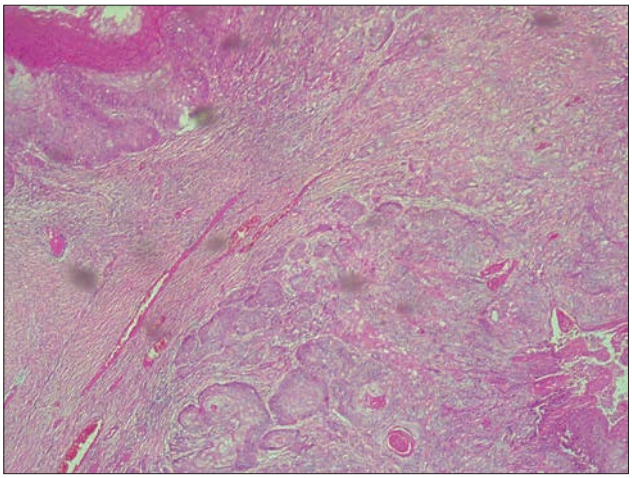

Figure 4: Histological section shows a cyst wall with an infiltrative well-differentiated tumor arrayed in nests, trabeculae, and small sheets, ×200

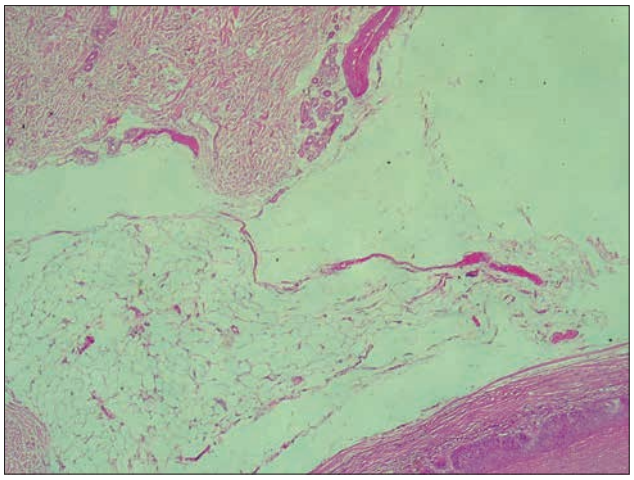

Figure 5: Section showing the deep resection margin that is uninvolved by the tumor

cytologic atypia, with numerous mitotic figures including abnormal ones. Other diagnostic tools include the use of fine needle aspiration cytology(FNAC),[12] though this may not be reliable or diagnostic. Immunohistochemistry using CD34 antibody marker has also been employed, but this is also not diagnostic by itself.[13]

There is no standardized treatment modality for malignant proliferating pilar tumor. Complete surgical excision may suffice in the absence of nodal or distant metastasis. The use of chemotherapy and/or radiotherapy has been advocated in tumors with metastasis, but it is still subjective and reliant on individual patient tumor extent. Mann *et al*. and some similar studies have reported that clinically aggressive tumors are associated with nodal or distant metastasis.[2,4,8,14] Our patient had surgical excision though the tumor extended to the margin. Incompletely excised tumors are fraught with recurrence and high risk of metastasis. There was no demonstrable nodal involvement in this patient as there were no palpable swellings or nodules. Patient has not agreed to another surgical exploration due to the diagnosis of incomplete excision.

Conclusion

Malignant proliferating trichilemmal tumor is rare and poses a diagnostic dilemma for the pathologist, especially in the resource-constraint environment. It has also been documented that the tumor has high tendency to recur especially if not completely excised and metastasize more frequently than squamous cell carcinoma.[11] Wide surgical excision should be considered as the first-line modality of treatment, while alternative therapies require further evaluation and individual health facility protocol.

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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