



Proliferating Trichilemmal Tumor: A Case Study

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Abstract

Proliferating trichilemmal tumor (PTT) is a scarce tumor of exterior root sheath that appears to grow within the wall of a pre-existing pilar cyst in most cases. This report presents a case of malignant PTT in the posterior sacrum of a 33-year-old man presenting with limited mobility and a history of paraplegia.

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Introduction

Proliferating trichilemmal tumor (PTT) is an infrequent tumor of exterior root sheath, which is classified into benign, locally invasive, and malignant categories (1). This type of tumor, which was first reported in 1966 by Wilson-Jones (2), is common on the scalps of older women over 60 years old. The malignant type of tumor is much rarer compared to the benign type and is characterized by trichilemmal keratinization (3). The main treatment for this type of tumor in cases of definitive malignancy is complete excision of the tumor while maintaining an appropriate margin to prevent its recurrence (4). This research reports a case of malignant PTT in the posterior sacrum of a young man. Our case report is a man with a large mass in the sacrum with a definite diagnosis of malignant PTT at bedsore, which is an infrequent case, undergoing surgical treatment and continuous follow-up by the surgical team. The patient was long-term bed-ridden with a huge sacral pressure sore, which may have induced chronic inflammation, and finally had a malignant transformation. Clinically, the wound behaves like a Marjolin ulcer, which is a cutaneous malignancy that arises from previously injured skin, longstanding scars, and chronic wounds. Its early recognition followed by surgical resection, if possible, is of utmost importance (7).

Case Presentation

A 33-year-old man was presented with limited morbidity and a history of paraplegia. The patient suffered from a spinal cord injury due to falling from a height of

27 m, years ago. He had paraplegia (lower extremity paralysis), paraparesis, incontinence, impotence, limited mobility, and deformity of the lower extremity joints. He developed bedsore in the posterior sacrum 15 years ago and occasionally received topical medications. The wound progressively increased in size over 15 years, and purulent drainage came out from the wound (Figure 1).

The patient was referred to the surgical clinic of the hospital and was visited by a surgeon for a wound biopsy, and the PTT was reported on the histopathology. The patient was a candidate for complete surgical excision of a large mass in the posterior sacrum with a margin of the gluteal tissue. The size of the removed mass was about 3.5 × 14 × 15 cm, and a malignant PTT excision was reported in this case (Figure 2).

Two months after complete removal of the mass, the patient presented with a complaint of tumor recurrence. After a comprehensive examination and viewing of the requested MRI report, he underwent surgery to remove the tail and the rest of the mass (Figure 3).

S2 and S3 vertebrae involvement in the sacral spine and invasion of the spinal canal were reported based on the secondary MRI results. Therefore, the sacrum between S5 and S2 had to be removed entirely and partially from the S1 vertebrae (Figure 4).

Usually, the removal of the S1 and S2 vertebrae is not performed since, if it is done, it can lead to fecal incontinence, urination, and impotence. These problems already existed due to our patient's previous spinal cord injury. Every time the patient refers to the clinic, suspicious

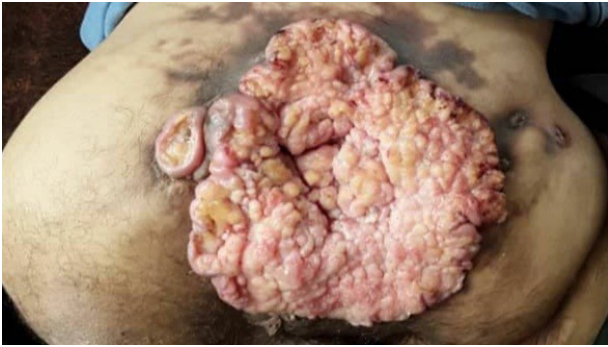


Figure 1. Initial Appearance of the Wound Before Sampling.



Figure 2. Complete Removal of the Mass Along With the Soft Tissue of the Guttural Area.



Figure 3. Tumor Recurrence Two Months After Mass Removal.

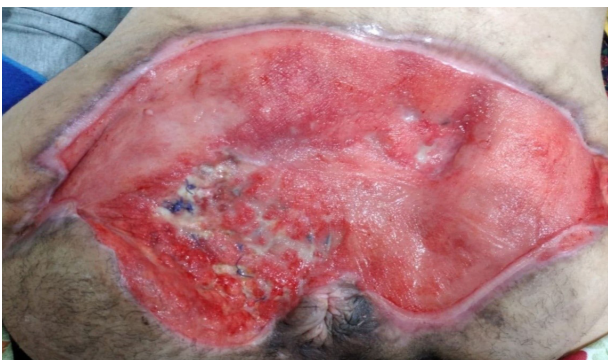


Figure 4. Three Weeks After the Removal of the Sacrum.

areas are burned with an infrared coagulator, mostly used to treat and reduce hemorrhoid complications. At present, the patient has been free from recurrence at a one-year follow-up (Figure 5).

Discussion

PTT is an uncommon but morphologically distinctive cyst usually occurring in the scalp of older women (1). These tumors are unlikely to develop in lanugo hair follicles on a hairless scalp or in follicles in other places devoid of nonterminal hair (4). They belong to the category of solid cystic neoplasm, which can occasionally progress from an adenomatous stage of the trichilemmal cyst to an epitheliomatous stage of the PTT, eventually leading to a carcinomatous stage of the MPTT (5). The cause of these tumors remains a mystery; in some reports, human papillomavirus, chronic inflammation, and sunlight are the causes of ulcers (6).

MPTT typically arises as a subepidermal tumor in women over 60 years of age, while the present case report highlights its occurrence in a young man. Our patient reported a large mass in the posterior sacrum with a margin of the gluteal tissue on the histopathology reporting. According to our patient's condition, bedsores can also be related to one of the predisposing factors for PTT. By searching authoritative study sources in the literature, no report was found about this type of malignant tumor in the posterior sacrum with a large area and invasion of the sacrum bones. In our study, the color of PTT-induced wounds was white to milky iodine, while in other published articles, the color of the wound varied between white and black (7).

There are different explanations for the treatment and behavior of PTT. Shetty et al reported scalp PTTs that were successfully excised with wide local excision and a 1-cm margin (8). In another study, Saida et al reported a patient with multiple trichilemmal cysts that were excised with a wide margin but later presented with metastases in regional lymph nodes with the same histological findings,



Figure 5. Wound Condition One Year After the Surgery.

suggesting that the original trichilemmal cysts may have been misdiagnosed and were PTTs (8). In this group of tumors, neoadjuvant treatment is also not performed because it significantly weakens the wound healing process, and resection surgery is recommended as much as possible (9). The most valid method is to remove the mass while maintaining a margin of 0.5 cm from healthy tissues (10).

Conclusion

MPTT is a rare malignant lesion and poses a diagnostic dilemma. The first step in diagnosing malignant tumors is a timely diagnosis of the type of the tumor. Therefore, accuracy in clinical and pathological results is highly important in differentiating malignant from benign. Although wide surgical excision should be considered the primary treatment modality, complex complications and problems after malignant tumor surgery, including tumor recurrence, can be prevented by long-term follow-ups.

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Authors' Contribution

NM and MRP: Conceptualization, the original draft writing, and investigation; FH: Reporting of the disease administration; NM: Writing, including reviewing and editing, and investigation.

Conflict of Interest Disclosures

There is no conflict of interests.

Consent for Publication

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

Disclaimer

The views expressed here are those of the authors and do not

necessarily reflect the views of the Ministry of Health and Medical Education.

Ethical Statement

This study was approved by the Ethics Committee of Urmia University of Medical Sciences with the code of IR.UMSU.REC: 1400.391.

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