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Case Report

Rare Site of Malignant Transformation: A Case Report

*Kumar A, Chowdhury P, Pratap A, Bhartiya SK, Saroj SK and Shukla VK**

Department of General Surgery, Banaras Hindu University, India

Abstract

Pilonidal sinus is a chronic disease which can affect many regions of the body, but particularly seen in the sacrococcygeal region in the natal cleft. Symptoms can range from mild scanty seropurulent discharge or in certain rare cases can also undergo malignant transformation. In this case, a 50 years old lady underwent malignant transformation in a pre-existing neglected pilonidal sinus.

Introduction

Pilonidal sinus is a common disease entity arising from multiple regions most commonly being sacrococcygeal region owing to its pathophysiology as mentioned by Karydakis [1]. Cellulitis, abscess growth, and recurrent sinus tract after surgery in the sacrococcygeal region are symptoms of this chronic inflammatory illness. A rare disorder is pilonidal sinus malignancy. The histological variant that occurs most frequently is squamous cell carcinoma. The preferred course of treatment for cancer involves wide local excision with substantial tumour free margin coupled with reconstruction, condition managed either by local or distant followed by adjuvant radiotherapy.

Case Presentation

A 50-years old lady presented with complaints of growth in the natal cleft from last 6 years. She had complained of discharge from a small opening in natal cleft from last 15 years which over the years developed into a fungating growth. At presentation, 10 cm × 8 cm fungating mass were present over the sacrococcygeal region. Tumor was overhanging over anal region although margin of the tumor was 2 cm away from anal verge. No ulcer or bleeding from the mass (Figure 1). Surrounding skin was normal. On digital rectal examination, anal tone was normal and no growth was present. No lymph nodes were palpable. Incisionl biopsy was done and reported as well differentiated squamous cell carcinoma. MRI pelvis and lower abdomen revealed left deep inguinal lymph nodes of size ~1.2 cm × 0.9 cm for which FNAC was done and was found as reactive lymph nodes. The anal sphincter and the anal verge were free of tumor. Wide local excision of the tumor (Figure 2) was performed and wound was left open and dressing was done with 1 cycle of NPWT application. Histopathology confirmed well-differentiated squamous cell carcinoma (Figure 3 and 4) with margins free of tumor with hair in core of lesion found during grossing. The patient received adjuvant radiotherapy and was referred to Department of Plastic surgery for reconstruction. There was a history of small lesion present in natal cleft with occasional discharge from six year before the appearance of present condition.

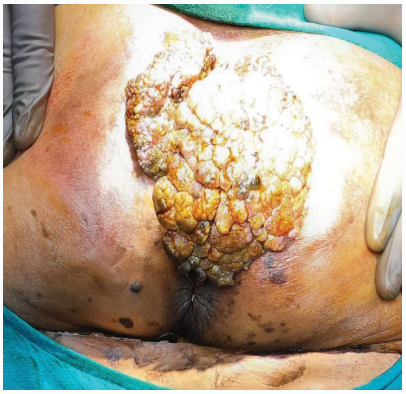

Figure 1: Pre-operative image of tumor over sacrococcygeal region.

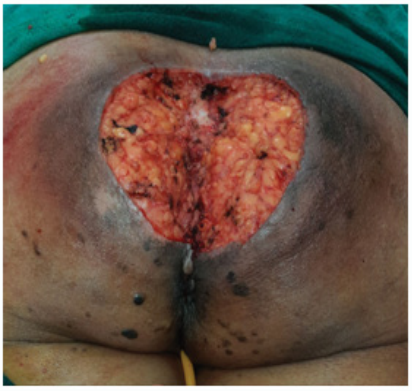

Figure 2: wound after wide local excision of tumor.

Discussion

Pilonidal illness is characterized by a sinus or abscess that contains hair and is frequently found in the gluteal cleft, also known as the natal cleft. Men are four times more affected than women, common in young adult [2]. Cellulites and abscess are the typical presentations of pilonidal illness [3]. Malignant transformation of pilonidal sinus is not more than 0.1 percent, the common variant being SCC. It is a dreadful long-standing complication with the average age at diagnosis is 52 years, and typically lasts 20 years before developing malignant changes [4,5]. Arising from any other chronic wound like that of protracted infection, such as chronic osteomyelitis or fistula-in-ano. Inflammation of the skin and persistent infection are the main

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***Corresponding author:** Shukla VK, Department of General Surgery, Banaras Hindu University, Varanasi, 221 005, India, Tel: +91-542-2309512; Fax: +91-542-2368163, 2367568

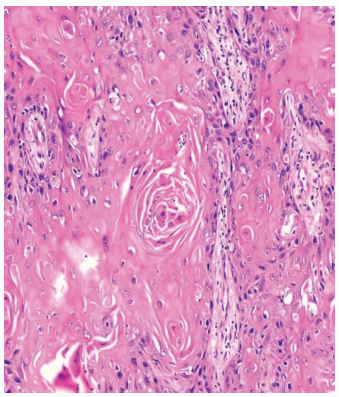

Figure 3: HPE reveals well differentiated squamous cell carcinoma.

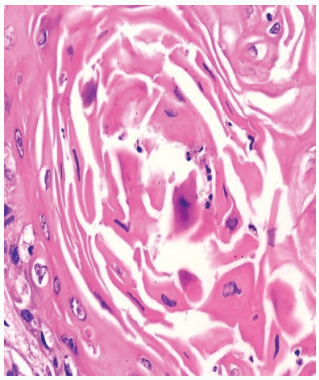

Figure 4: HPE reveals well differentiated squamous cell carcinoma.

etiological factors. A developing, ulcerated tumor with indurated borders raises clinical suspicions of pilonidal cancer [6]. In present case, a 50-year-old female presented on outpatient basis with 6 years history of painless progressive fungating mass in the sacrococcygeal region on further enquiry she recalled discharging sinus in the gluteal fold for last 15 years.

Pilonidal cancer has a characteristic look, and a physical examination is typically sufficient to make the diagnosis. With a friable, indurated, erythematous, and fungating edge, a core ulceration is frequently seen, not observed in our case. Nodal metastases occur in 14% of cases when there are also inguinal lymph node metastases [7,8]. Although there were clinically impalpable inguinal lymph nodes in our case, MRI pelvis and lower abdomen revealed enlarged left deep inguinal node which turned out to be reactive on USG guided FNAC.

If broad tumor-free margins are not created during initial surgery, the 5-year survival rate falls to 55% from the current overall rate of 61% [8]. In individuals who presented with nodal metastases, Pilipshen et al. reported that there were no survivors at 2 years [8]. It is uncommon to survive for ten years [9], and metastatic disease is frequently described [5,10].

Ideal is to perform an intraoperative biopsy of the margins of the resected tissue but in this case, covering of the defect was postponed as sufficient intraoperative histological analysis was not achieved. The defect's reconstruction in our case was done using delayed fascio-cutaneous flap by the plastic surgeon team. Primary wound closure after 1 cycle of NPWT in the same setting is an option for post excision small size wound. For the reconstruction, either skin grafts or local flaps can be employed, depending on the size of the postoperative lesion. Free tissue transfer (using muscular or musculocutaneous microsurgical flaps) is another option [11].

The optimum course of action for the first management of neoplastic degeneration of Pilonidal Sinus is surgical removal of the main tumor with disease free margins [12]. This applies to the gluteal muscles, sacrum, and coccyx if the tumor has spread to those areas. If the pelvic floor is involved by rectal infiltration, it may also be essential to conduct an abdominoperineal amputation 19 or perhaps a hemicorporectomy or Tran's lumbar amputation [13]. Adjuvant chemotherapy and radiation have been suggested by several authors as a way to reduce the rate of local recurrence. Recurrence rates drop from 44% to 30% when radiation is added to surgery alone [7]. Some authors advise giving radiation therapy in all instances following tumor removal [14,15].

After receiving adjuvant radiation therapy, this patient was followed up regularly and was found to be tumor-free for the following 8 months. Regular follow-up and surveillance are essential to detect any recurrence or metastasis. Further research is needed to understand better the pathogenesis and optimal management strategies for pilonidal sinus malignancy.

Conclusion

This report highlights a rare occurrence of malignant transformation in a neglected pilonidal sinus. Pilonidal sinus malignancy as squamous cell carcinoma. Timely diagnosis and appropriate management are crucial in improving patient outcomes. Wide local excision (R0) followed by reconstruction is the preferred. Adjuvant chemo radiotherapy may be considered to reduce the risk of local recurrence.

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