

A Rare Case of Squamous Cell Carcinoma in Chronic Sacrococcygeal Pilonidal Disease

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Abstract: Squamous cell carcinoma (SCC) arising in a chronic pilonidal sinus is a rare but serious complication, with an estimated incidence of 0.1%. This case report presents a 25 - year - old male diagnosed with well - differentiated SCC in the sacrococcygeal region, highlighting the clinical presentation, diagnostic challenges, and surgical management. The case underscores the importance of histopathological examination of long - standing pilonidal sinus lesions to detect malignancy early.

Keywords: Pilonidal sinus, Squamous cell carcinoma, Sacrococcygeal, Chronic disease, Case report

1. Introduction

Pilonidal sinus disease is a chronic inflammatory condition characterized by a tract or cyst containing hair, often occurring in the sacrococcygeal region. It is most commonly seen in young adults, particularly males between the ages of 20 and 30, and is often attributed to excessive sitting or local trauma, which leads to ingrown hair in the natal cleft. The condition is typically benign but can become recurrent and complicated, requiring repeated incision and drainage (1).

Malignant transformation of pilonidal sinus into squamous cell carcinoma (SCC) is exceedingly rare, with a reported incidence of about 0.1% in chronic cases (2). Despite its rarity, SCC in pilonidal sinus disease poses significant diagnostic and therapeutic challenges due to its aggressive nature.

Aims and Objectives

Our case report discusses a rare instance of SCC in a chronic pilonidal sinus in a young male patient, illustrating the clinical features, diagnostic process, and surgical intervention.

2. Review of Literature

Pilonidal disease is a gluteal cleft infection that occurs beneath the skin. In literal terms, pilonidal means "nest of hair." An estimated 26 out of 100,000 people are affected, with a 3:1 male predisposition (1). Obesity, local trauma, sedentary work, and family history are recognized risk factors. The condition was believed to be congenital because of the failure of fusion in the dorsal midline, which resulted in entrapment of hair follicles in the sacrococcygeal region. However, recent studies strongly support an acquired etiology (2). Interestingly, occurrence of pilonidal disease in the interdigital space of sheep shearers, dog groomers, cow milkers, and barbers further support the acquired etiology (1).

There are several proposed theories to explain the etiology of pilonidal disease. Bascom has noted that, inflammation of hair follicles with keratin results in local infection and abscess formation while suction causes hairs to enter and lodge in

abscess cavity. However, hair follicles are rarely seen in pathological examination (2). The cavity mainly contains hair, debris and granulation tissue.

Karydakis explains that the impaling of loose hairs causes a foreign body reaction. Subcutaneous burrowing of hairs result in secondary pit formation and additional hairs entering the through the pits. Pathogenic formula of Karydakis has three primary variables namely, loose hair (H), force (F), vulnerability of local skin (V). Force of insertion of hair (F) is influenced by factors like depth, narrowness and friction of the natal cleft (1).

3. Materials and Methods

A 25 - year - old male presented to the Institute of General Surgery with a six - month history of an ulceroproliferative lesion in the sacrococcygeal region. The patient reported a long - standing history of pilonidal sinus disease, initially diagnosed at the age of 19. Over the years, he had undergone multiple incision and drainage procedures for recurrent abscesses in the same region and received several courses of antibiotics. However, his symptoms never resolved completely, with persistent discomfort and occasional discharge. Upon examination, the patient had a large, 7 × 5 cm ulceroproliferative lesion in the sacrococcygeal area (Figure 1).



Figure 1: Large ulceroproliferative lesion of 7×5 cm in sacrococcygeal region.

The lesion was friable with a foul - smelling, seropurulent discharge, and surrounding induration. Systemic examination revealed no significant findings, and the patient denied any systemic symptoms such as fever or weight loss. The inguinal lymph nodes were not palpable, suggesting the absence of regional metastasis at that stage. Digital rectal examination was normal, with no evidence of deeper tissue invasion. Imaging studies, including a contrast - enhanced computed tomography (CECT) scan of the pelvis and magnetic resonance imaging (MRI) of the lumbosacral spine, revealed a 4 × 3 × 6 cm mass extending to 1.3 cm posterior to the S5 vertebra. The lesion spanned from the lower border of S2 to the sacrococcygeal region.

Positron emission tomography - computed tomography (PET - CT) confirmed the absence of distant metastasis. A biopsy of the lesion was performed, and histopathological examination revealed well - differentiated SCC. Swab culture of the wound identified *Staphylococcus aureus*, sensitive to imipenem, amoxicillin, and clavulanic acid. Based on these findings, a diagnosis of SCC arising in a chronic pilonidal sinus was confirmed. The patient was presented in the multidisciplinary tumour board and was suggested upfront surgery. The patient underwent a wide local excision of the lesion with a rotational flap closure to ensure adequate margins and reduce the risk of recurrence (Fig 4, 5, 6, 7).



Figure 2: MRI scan of the lumbosacral spine showing the tumor mass extending from the lower border of S2 to the sacrococcygeal region.



Figure 3: PET - CT showing increased FDG uptake in a heterogeneously enhancing irregular nodular lesion in intergluteal cleft.

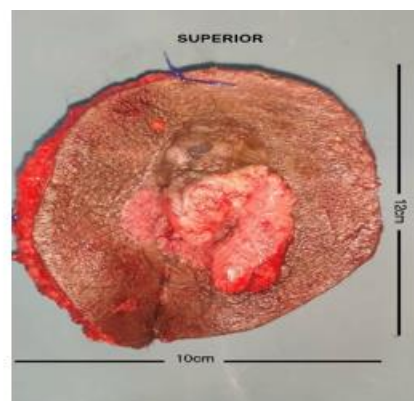


Figure 4 (Top left), 5 (Top Right), 6 (Bottom left), 7 (Bottom right) showing Clinical picture, Post Excision Raw area, Post Rotational Flap closure and Specimen picture respectively

4. Results and Follow up:

The excised specimen was sent for further histopathological analysis, which confirmed the findings of well differentiated

squamous cells carcinoma with clear resection margins and closest of 1.8cm in depth. The patient was represented in multidisciplinary tumour board and was advised to be discharge with regular 3monthly follow ups to monitor for

signs of local recurrence. Postoperatively, the patient recovered well without any immediate complications. Patient has been followed up for 1 year now and has no signs of recurrence or new lesions, with a healthy flap.

5. Discussion

Pilonidal sinus disease is an acquired condition often referred to as "jeep - bottom" due to its prevalence among soldiers and individuals who spend prolonged periods sitting. It typically presents with abscess formation, cellulitis, or persistent drainage from the sinus tract. However, the transformation into malignancy is rare and tends to occur in cases with chronic, untreated disease (3). The pathogenesis of SCC in a pilonidal sinus is not fully understood, but chronic inflammation, persistent infection, and repeated trauma to the area are considered significant risk factors. These factors may contribute to squamous metaplasia of the sinus epithelium, eventually leading to malignant transformation (3). Previous studies, such as those by Katsikeris and Kakarantza - Angelopoulou (2017), have reported similar cases, emphasizing the need for vigilance in patients with long - standing disease (4).

SCC arising from pilonidal sinus typically presents as an ulcerative or proliferative lesion with foul - smelling discharge, as observed in our patient. The diagnosis is often delayed due to the nonspecific nature of the symptoms and the rarity of malignancy in pilonidal disease. Imaging techniques like MRI and PET - CT are useful for assessing the extent of local invasion and ruling out metastasis, which is critical for surgical planning (5). The primary treatment for SCC in pilonidal sinus is wide local excision, aiming to achieve clear surgical margins to prevent local recurrence. In some cases, adjuvant radiotherapy may be considered for patients with incomplete resection or positive margins (6). However, in well - differentiated cases with complete resection, surgery alone is often sufficient. Gönenç et al. (2010) reported similar outcomes in patients treated with wide excision, underscoring the importance of early detection and aggressive surgical management (5).

Postoperative care includes regular monitoring for recurrence through physical examinations and, when necessary, imaging studies (4). Follow - up care is essential due to the potential risk of recurrence, even after achieving clear margins. The overall prognosis depends on the tumor grade, size, and presence of lymph node involvement or distant metastasis.

6. Summary and Conclusion

Malignant transformation of pilonidal sinus into SCC, though rare, is a serious complication that requires prompt recognition and intervention. This case highlights the importance of considering malignancy in patients with long - standing pilonidal sinus disease, especially those with recurrent infections or non - healing lesions. Routine histopathological examination of excised pilonidal sinus tissue can aid in early diagnosis and improve patient outcomes.

In this case, wide local excision with rotational flap closure provided a favorable outcome, with no evidence of recurrence on follow - up. This emphasizes the need for early surgical

intervention and a multidisciplinary approach in managing complex cases of pilonidal sinus with potential malignancy.

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