



Case Report

Squamous Cell Carcinoma Arising in Recurrent Pilonidal Sinus in an Older Woman

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Abstract

Malignant transformation is a rare complication of pilonidal sinus. We report the case of a 90-year-old female who had a recurrent pilonidal sinus with fistulous abscess formation. We excised the whole pilonidal sinus because the infection could not be controlled by oral antibiotics. Squamous cell carcinoma (SCC) was found in the sinus tract with a negative surgical margin. A whole abdominal computed tomography scan was performed, which showed no distant metastasis and no enlargement of the bilateral inguinal lymph nodes. This case study illustrates that SCC transformation may present in the recurrent and long-standing pilonidal sinus. We suggested that all pilonidal sinus lesions should be sent for pathologic examination to early detect malignant transformation.

Keywords: Pilonidal sinus, Squamous cell carcinoma, Malignant transformation

INTRODUCTION

Pilonidal disease is very common and is often complicated by infection. It occurs four times more frequently in men than in women.^[1] Malignant transformation is rare and occurs in approximately 0.1% of these patients, and mostly presents at an advanced stage or as incidental findings. The most common type of malignancy is squamous cell carcinoma (SCC).^[2] We report the case of a 90-year-old female in whom early SCC was found incidentally when excising a recurrent pilonidal sinus with fistulous abscess formation.

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CASE REPORT

A 90-year-old female patient denied any systemic diseases, but with a history of pilonidal cyst surgery more than 10 years previously at another hospital. She came to our clinic because of recurrent fistula formation with purulent discharge for several days. A recurrent pilonidal sinus infection with abscess formation and local cellulitis was suspected, and she was admitted for intravenous antibiotic treatment. A bacterial culture from the purulent discharge showed *Proteus mirabilis*. However, the infection recurred after stopping the antibiotics,

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and she underwent surgery to remove all fistulous abscess tracts. Surgery consisted of wide excision of all pilonidal sinus tracts deep to the gluteal muscle fascia, presacral fascia, and ischiorectal fossa. The wound was approximately 12 cm × 9 cm in diameter and 4 cm in depth. The wound was not closed, and the margin was marsupialized with a running suture. No flap or skin graft was used because of the initial wound infection, and the patient did not want to undergo surgery again because of her age and high risk posed by anesthesia. The wound healed by secondary healing over approximately 6 months [Figure 1].

A pathological study revealed a pilonidal sinus with a sinus tract lined by acanthotic squamous epithelium and surrounded by dense acute and chronic inflammatory cells, granulation tissue, and fibrosis. Foci of SCC arising from the sinus tract were noted and the section margin was free of malignancy [Figure 2]. A whole abdominal computed tomography (CT) scan was performed, which showed no distant metastasis and no enlargement of the bilateral inguinal lymph nodes. No tumor recurrence was noted after 1 year of follow-up.

DISCUSSION

Pilonidal disease occurs because of a skin infection, mostly at the crease of the buttocks or natal cleft. The most accepted pathogenesis involves penetration of hair follicles into the skin, which induces a foreign body reaction and infection.^[3] Untreated pilonidal disease may result in complications such as recurrent abscess formation, local cellulitis, and less frequently, sacral osteomyelitis or meningitis. Several surgical treatments have been reported in the literature, including simple incision and drainage, total sinus excision with primary or secondary intention method closure, marsupialization, and flap closure.^[4]

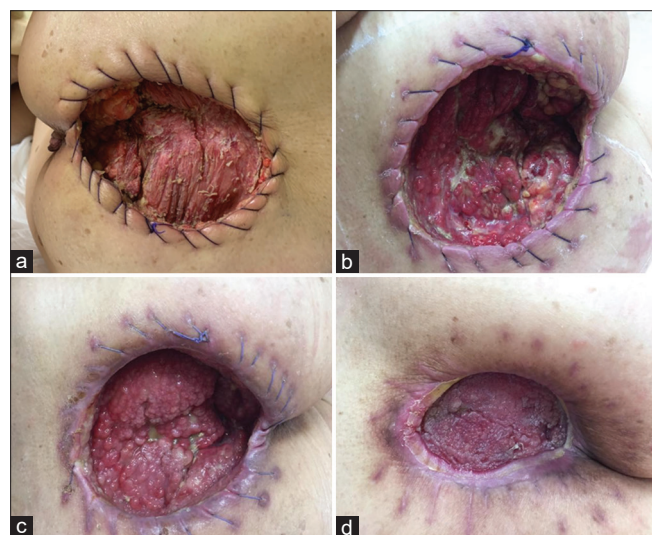


Figure 1: Postoperative wound healing. (a) Three days after the operation, showing the bilateral gluteal muscles, presacral fascia, and anococcygeal ligament. (b) Two weeks after surgery. (c) Five weeks after surgery. (d) Three months after surgery

Wolf described the first case of SCC arising in a pilonidal sinus in a 21-year-old woman in 1900,^[2] and fewer than 100 cases of pilonidal malignancies have been reported in the literature since then.^[4] The incidence of malignant degeneration is 0.1%, with an untreated disease duration of 20–30 years or with recurrent pilonidal sinus. Around 70% of the patients who develop carcinoma from a pilonidal sinus have had symptoms for more than 10 years.^[5] The mechanism leading to malignant degeneration seems to be related to a compromise of DNA-repair mechanisms due to chronic inflammation, which causes the release of free oxygen radicals by activated inflammatory cells.^[4]

SCC is the most common carcinoma associated with chronic pilonidal disease, accounting for approximately 88% of these patients. Other types of malignancies, such as basal cell carcinoma and sweat gland adenocarcinoma, have also been reported.^[2] The tumor tends to grow slowly, but has an aggressive tendency toward the local invasion of the anus, rectum, or sacrum or coccyx bone involvement. Pilonidal carcinoma has a distinctive appearance, with the diagnosis frequently being made on inspection. It can be detected incidentally on postexcisional histopathology. A central ulceration is often present, with a friable, indurated, erythematous, and fungating margin in advanced disease.^[6] Inguinal nodal metastasis has been reported in about 14% of cases. A physical examination of the inguinal area or a CT scan can detect enlarged lymph nodes, and a fine-needle aspiration biopsy can provide pathological proof.^[3] The preoperative evaluation also needs to include sigmoidoscopy and pelvic magnetic resonance imaging. These procedures can evaluate the depth of the tumor invasion and detect any rectum/anus involvement preoperatively. Positron emission tomography is a useful modality to identify metastasis.^[7]

En bloc wide excision is suggested if there is no inguinal lymph node involvement or distant metastasis. The clinically healthy tissue margin must be at least 3 cm. The sacral fascia needs to be removed, and coccygeal excision and decortication or resection of the sacrum must be performed if there is bone infiltration on a CT scan.^[8] Although presentation with inguinal adenopathy is a poor prognostic sign,^[1] prophylactic inguinal node dissection is not recommended because it does

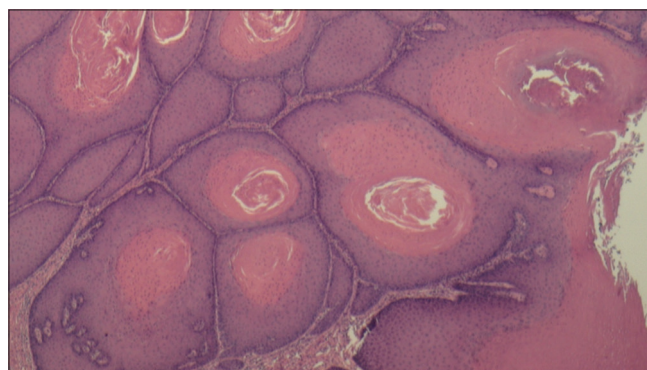


Figure 2: Foci of squamous cell carcinoma arising from the sinus tract

not improve the survival of patients with occult metastasis.^[8] Almeida-Goncalves presented a series of seven cases of SCC in pilonidal sinuses that were considered to be inoperable and were treated with liquid nitrogen cryotherapy, with a complete response in 100% of the patients.^[9]

The overall recurrence rate, including local recurrence and metastasis, ranges from 44% to 50%. Wide excision with tumor-free margins has reportedly resulted in a 5-year disease-free survival rate of 55% and overall 5-year survival rate of 61%.^[3] Local recurrence may be decreased to 30% with adjuvant radiotherapy.^[1] The role of adjuvant chemotherapy remains unclear, but some reports have shown that it may be effective in combination with resection and radiotherapy for high-risk lesions.^[7] There are currently no reports on the use of neoadjuvant concurrent chemoradiotherapy in advanced cases, but neoadjuvant radiotherapy was suggested by de Bree *et al.*, because it is expected to reduce the viability of tumor cells that may seed in the operative field, and this may reduce the size of the tumor.^[1]

CONCLUSION

Malignant degeneration is a rare but serious condition in chronic inflammatory pilonidal disease. Every excised specimen should be examined histologically, especially in older patients (>40 years old) with long-standing disease.^[4] Wide excision with a tumor-free margin is the only way to cure early malignancy in pilonidal disease, but the early surgical treatment of the pilonidal disease itself is the best way to prevent malignant degeneration.^[5]

Declaration of patient consent

The author certifies that he has obtained all appropriate patient consent forms. In the form, the patient has given

her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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