Squamous cell carcinoma arising from pilonidal sinus

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Key words
Cancer; Sinus; Natal cleft; Sacrococcygeal region

Abstract
Pilonidal sinus is usually present in the sacrococcygeal region. The common presentations are cellulitis, abscess or sinus. Rarely malignant change may be seen in chronic pilonidal sinus. We report a case of chronic pilonidal sinus complicated with squamous cell carcinoma.

Introduction
Pilonidal sinus is a chronic inflammatory condition characterised by cellulites, abscess formation and recurrent sinus tract in the sacrococcygeal region. Malignancy in pilonidal sinus is a rare condition. Squamous cell carcinoma is the most frequent histological type. Wide excision is the treatment of choice for carcinoma associated with pilonidal sinus with closure by rotation flap or skin grafting. We report a patient with squamous cell carcinoma developing on a chronic pilonidal sinus of 19 years duration.

Case report
A 58-year-old male was admitted to the University Hospital of Banaras Hindu University, India, with a large ulceroproliferative lesion in the sacrococcygeal region. After detailed inquiry, the patient gave a history of pilonidal sinus which was diagnosed at the age of 39 by a specialist surgeon outside this hospital. He refused the offered surgical treatment. Instead, he underwent repeated incision and drainage and received multiple courses of antibiotics for the abscess. The disease was never cured completely.

Six months ago, an ulceroproliferative growth appeared with foul smelling seropurulent discharge.

Systemic examination was unremarkable. A large 13 × 13 cm ulceroproliferative friable lesion was present on the sacrococcygeal region up to the sacral bone with surrounding induration (Figure 1). The inguinal lymph nodes were not enlarged. Digital rectal examination was normal. Laboratory analysis revealed haemoglobin level as 9 gm/dl, WBC count was 14 500/cumm and ESR 41 mm/hour.

A CT scan of the pelvis and X-rays of lumbosacral spine and pelvis were normal. USG abdomen and X-ray chest were also normal. Swab culture of the wound revealed Staphylococcus aureus that was sensitive to imipenem, amoxycillin and clavulanic acid.

Preoperative biopsy was taken from multiple sites of the lesion from the margins, which was suggestive of moderately differentiated squamous cell carcinoma. Wide excision of large tumour was done and the defect was closed with a

Key Messages
• in long standing pilonidal sinus with ulceration, malignancy should be kept in mind
gluteal skin flap. The specimen histology was also moderately differentiated squamous cell carcinoma (Figure 2). Resected margins were clear. No further treatment was given to the patient. The patient has been on regular follow-up for the past 2 years with no evidence of recurrence.

Discussion

Pilonidal disease consists of a hair-containing sinus or abscess occurring most frequently in intergluteal cleft, also called the natal cleft. The disease occurs primarily in young adults and is four times more common in men (1). The most common presentation of pilonidal disease is cellultes and abscess (2).

This disease is generally benign and malignancy is extremely rare in this setting. However, a few cases have been reported worldwide with different types of malignancy, most of which have been squamous cell carcinoma. Other reported malignancies are basal cell carcinoma and adenocarcinoma of sweat gland type (3).

The incidence of carcinoma arising in pilonidal disease is less than 0.1% (4). In our case, the patient was a 58-year-old male, and it took 19 years to develop malignant changes. Carcinoma in pilonidal sinus wounds occurs more often in males, and the average age at diagnosis is 52 years and the mean duration of pilonidal disease before malignant alteration is 20 years (5). Any long-standing infection as fistula in ano, chronic osteomyelitis, etc. can develop squamous cell carcinoma similar to pilonidal sinus. The most important aetiological factor is chronic infection and skin irritation. Pilonidal carcinoma is suspected clinically by growing, ulcerated mass with indurated edges.

The tumours arising from pilonidal disease tend to be slow growing. However, many have a tendency toward aggressive local invasion and metastasis (3,6). Ulceration, overgrowth, seropurulent drainage and inguinal adenopathy are gross symptoms suggesting malignant association. Presentation with inguinal adenopathy is a poor prognostic sign (3,7).

Diagnosis is confirmed by biopsy. The most important consideration in the histologic differential diagnosis of pilonidal carcinoma is pseudocarcinomatous hyperplasia of squamous epithelium, which may in fact be in association with any severe inflammatory process (8).

Surgical procedure includes wide excision of the healthy tissue including skin, subcutaneous tissue and gluteal muscle by a 3-cm margin. Sacral fascia should be removed and sometimes coccygeal excision and decortication or resection of the sacrum is needed to be performed if bone infiltration is seen on CT scan (9). Prophylactic inguinal node dissections are not recommended, but groin dissections must be performed if there is presence of positive lymph nodes.

Reconstruction of the defect could be performed primarily or delayed. Depending on the size of the postoperative defect either skin graft or local flaps are being used for the reconstruction. Also free tissue transfer (microsurgical flaps either muscular or musculocutaneous) can be applied (9). In our case, the postoperative defect was closed with local skin flap.

Some authors propose consideration of adjuvant chemotherapy and radiation to decrease the local recurrence rate. When radiotherapy is added to surgery alone, recurrence rates decrease from 44% to 30%. A 10-year survival rate is rare (3), and metastatic disease is often described (5,9). Re-excision of local recurrence resulted in some long-term survivals (9,10).

Conclusion

The incidence of malignancy is far too small for pilonidal sinus to be considered as carrying a significant malignant potential. However, in long-standing pilonidal sinus, the presence of a hidden carcinoma should be suspected. All excised specimens of pilonidal sinus lesions should be sent for pathologic examination. The best prevention to avoid malignancy is surgical treatment of pilonidal disease in its early stage.

References


