

Verrucous squamous cell carcinoma arising in sacrococcygeal recurrent pilonidal sinus: A case report

Sakrokoksigeal rekürren pilonidal sinus kaynaklı verrüköz skuamöz hücreli karsinom: Bir olgu sunumu

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Abstract

Pilonidal sinus is an acquired and well described disease which is mostly located in sacrococcygeal midline region, lined by squamous epithelium and characterized by chronic inflammation including foreign body giant cells and sinus or sinuses containing epithelial debris and sometimes young granulation tissue. A malignant transformation can be seen in 0.1% of chronic or recurrent pilonidal sinus disease. The present case was 63 years old male patient undergoing pilonidal sinus operation 35 years ago. In the physical examination, there was an ulcerovegetating mass of 10 cm in diameter, including multiple orifices of sinuses that contained purulent material drained by squeezing. The histopathological examination of the mass resected with wide excision revealed a typical verrucous squamous cell carcinoma. There were no lymphovascular and perineural invasions, also any distant metastases was observed in this case.

Keywords: Pilonidal sinus, squamous cell carcinoma, carcinomatous transformation

Özet

Pilonidal sinüs, sakrokoksigeal orta hat lokalizasyonlu, skuamöz epitelle döşeli, yabancı cisim devhücreleri içeren kronik inflamasyon tablosu, epitelyal debris ve bazen genç granülasyon dokusu içeren sinüs veya sinüslerle karakterize iyi tanımlanmış edinsel bir hastalıktır. Kronik veya rekürren pilonidal sinüs hastalığında malign transformasyon yaklaşık %0,1 oranında görülebilir. Olgumuz 35 yıl önce pilonidal sinüs operasyonu geçiren 63 yaşındaki erkek hastadır. Fizik muayenede basmakla püy içeriği boşalan sinüslere ait multipl açıklıklar içeren 10 cm çapındaki ülserovejetan görünümlü kitle izlendi. Geniş eksizyonla çıkarılan kitlenin histopatolojik incelemesinde tipik verrüköz skuamöz hücreli karsinom saptandı. Lenfovasküler ve perinöral invazyon izlenmeyen olguda uzak metastaz da saptanmadı.

Anahtar sözcükler: Pilonidal sinüs, skuamöz hücreli karsinom, karsinomatöz transformasyon

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Introduction

Pilonidal sinus is a common, acquired, inflammatory and well recognized disease which is mostly located in sacrococcygeal midline region, lined by squamous epithelium and characterized by sinuses containing chronic granulation tissue, hair follicles, epithelial debris and sometimes has young granulation tissue. Hodge used a term "pilonidal" (Latin: pilus=hair and nidus=nest) in 1880 and pilonidal sinus was first described as a hair containing sinus by Herbert Mayo in 1883 [1].

Sinuses and the tracts of sinuses are small openings located in midline region, lined by stratified squamous epithelium inside and infiltrated with many inflammatory cells including neutrophils, lymphocytes, plasma cells and sometimes haemosiderin-laden macrophages and also foreign body giant cells. Hair shafts can be seen in fifty to seventy-five (50-75%) of the cases, deeply in granulation or scar tissue, or lying free in the cavity of cysts [1, 2].

Pilonidal sinus can be symptomatic with pain, swelling and acute abscess formation, whereas the disease may also remain asymptomatic for many years. A big ratio of these abscesses recovers spontaneously but some cases require surgical resection. Chronic untreated pilonidal sinuses make an anastomosis to skin with multiple orifices and subcutaneous hair containing vesicles occur in years. The presence of these hairs in sinuses impairs wound healing and causes chronicity by chronic irritation and foreign body reaction [3].

In the presence of chronic or recurrent pilonidal sinus, there can be rarely (0.1%) a malignant transformation usually as squamous cell carcinoma and/or verrucous carcinoma [1, 4]. Cutaneous squamous cell carcinomas are often slow growing and painless tumors that can be nodular, ulcerous, plaque like or verrucous. Verrucous carcinoma is a low grade squamous cell carcinoma that is first reported inside oral cavity in 1948. At the beginning it is an exophytic, verrucous tumor, it can penetrate deeply in tissue [5].

In the present case, carcinomatous transformation in recurrent and persistent pilonidal sinus disease was reported with some literature.

Case report

Our case was 63 years old male patient who had pilonidal sinus operation 35 years ago, and in the following years, although there was a recurrence and persistent fistula tract, he refused treatment. Physical examination findings were inflammation on intergluteal sulcus, multiple orifices of sinuses that contained purulent material and ulcerovegetating mass of 10 cm in diameter, extending to left gluteal region. After the resection of the mass with wide excision, the tissue defect was covered with multiple skin flaps. The mass was macroscopically indurated as 2 cm from skin, 10 cm in diameter and had 5 cm thick, including the subcutaneous fatty tissues, fragile, villiform and keratinous (Figure 1).

In microscopic examination, a layer of thick parakeratosis and compact orthokeratosis on the surface, the histological appearance of squamous epithelium with minimal atypia as typical "verrucous squamous cell carcinoma" were observed (Figure 2). Tumor was distributed throughout the sinus tracts. Around the invasive tumoral nests there was dense lymphoplasmacytic infiltrate in large areas (Figure 3). There was no lymphovascular or perineural involvement, also any distant metastasis and/or lymph node metastasis was seen in this case. After the healing of flap, oncology consultation was performed.



Figure 1. The macroscopic appearance of the fragile tumor with villiform surface, invading the subcutaneous fatty tissue.

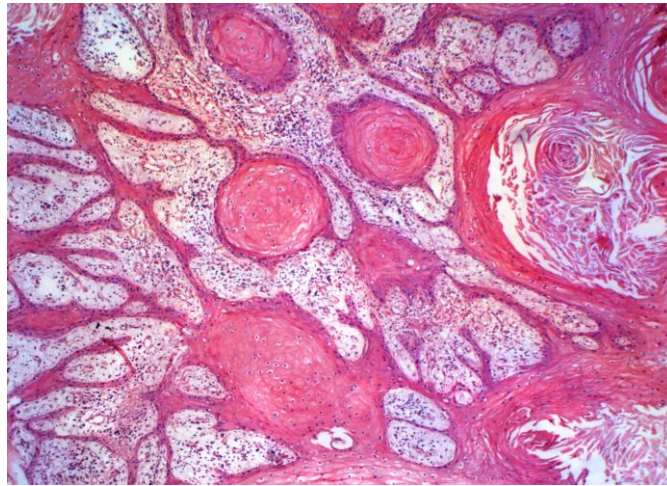


Figure 2. The areas of verrucous squamous cell carcinoma showing minimal atypia (H&Ex50).

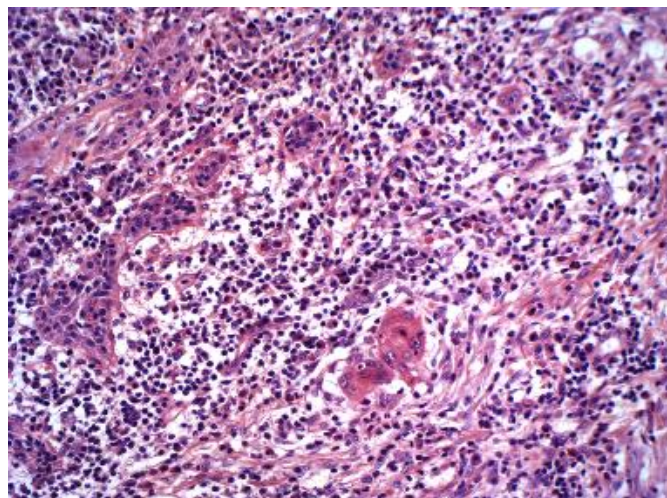


Figure 3. Islands of malignant squamous epithelium with minimal atypia and intense malignant epithelial cell groups around the tumor, lymphoplasmocytic chronic inflammatory cell infiltration (H&Ex200).

Discussion

Pilonidal sinus disease is usually seen in sacrococcygeal region and in men under the age of 40 [2]. In etiopathogenesis, local microtraumas, hormonal predisposition and hair density are accused and the involvement in interdigital spaces of barbers and sheep shearers who have usually microtraumas and sacrococcygeal region in soldiers and long distance drivers show the occupational predisposition [1, 3].

Disease sometimes remains as an asymptomatic sinus for years, or can be symptomatic with pain, swelling and acute abscess formation. Most of these abscesses heal spontaneously, a part of them requires a surgical resection. Recurrence is frequently observed in the cases of hair accumulation in the sinus if it isn't surgically fully cleaned. Also the cases that surviving acute abscess formation lighter, may present with the chronic draining complex sinus and fistula tracts.

Pilonidal sinus disease can be easily diagnosed on clinical findings and in these patients due to staying for a long time without any treatment transformation to squamous cell carcinoma can be seen as a rare complication [4]. In literature, 61 cases have been reported and these cases have a higher recurrence and poor prognosis than the other non-melanoma skin cancers [6, 7]. Although accepted treatment methods are wide resection and radiotherapy, local and regional recurrence rate is reported to be as high as 50% [7, 8].

The pathophysiology of carcinomatous transformation in pilonidal sinus disease is suggested due to disruption of DNA repairment by chronic inflammation and free oxygen radicals that released by inflammatory cells [7, 8].

Verrucous squamous cell carcinoma that diagnosed in our case, has markedly morbidity and nearly 70% of recurrence especially in anal and perianal lesions and has mortality rates ranging between 20-30%. Metastasis is rarely reported [9].

Therefore in elder patients as our case, especially in long-standing persistent pilonidal sinus diseases and in the presence of the adjacent atypical lesions, malign degeneration should be considered and it should be noted that treatment modality may require a wide excision of the mass, lymph node dissection and in order to prevent local recurrence postoperative radiotherapy can be performed addition to standard pilonidal sinus treatment.

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