Annals of Clinical Case Reports

Case Report
Published: 01 Aug, 2022

A Giant Squamous Cell Carcinoma in a Patient with Pilonidal Sinus: A Case Report

Talal Alshareef[1], Raief Alfriedy[1], Ruba Aldhaheri[2], Nahlah Arab[1] and Fatima Badahdah[1]*

[1]Department of General Surgery, Prince Sultan Military Medical City, Saudi Arabia

[2]College of Medicine, King Abdulaziz University, Saudi Arabia

Abstract

Background: Pilonidal disease is among the most common surgical problems, and inflammatory complications of the disease, such as cellulitis, abscesses, and recurrent sinus tracts, are well known to general surgeons. In rare cases, longstanding inflammation can lead to malignant transformation, most commonly to Squamous Cell Carcinoma (SCC), the treatment of choice for which is total surgical resection with free margins. Tissue defects can be repaired using rotational flaps or skin grafts.

Case Report: We describe an extremely rare case of SCC in a 66-year-old male retired truck driver with a 5-year history of a chronic discharge and a protruding mass measuring 6.5 cm × 14.2 cm × 16.8 cm on the midline in his gluteal region. The condition was diagnosed by punch biopsy. Preoperative staging with total-body computed tomography was performed to rule out metastasis. Finally, the patient underwent total surgical resection of the mass and coverage with a rotational locoregional flap, skin graft, and vacuum-assisted closure the postoperative course was uneventful and his condition improved markedly.

Conclusion: SCC arising in a pilonidal sinus is a rare complication of a common disease. The present case is extraordinary in many respects, since we paid attention to the tumor site and because the buttocks are not a sun-exposed site. Moreover, massive SCCs are accompanied by the risk of disfigurement, local recurrence, and metastasis. Finally, surgery is the mainstay of treatment even in giant tumors, and in some cases, it should be followed by radiotherapy to address the risk of metastatic spread.

Keywords: Pilonidal disease; Squamous cell carcinoma; Gluteal region; Flap

OPEN ACCESS

Correspondence:
Ruba Aldhaheri, College of Medicine, King Abdulaziz University, Jeddah, Saudi Arabia, Tel: +966556676101; E-mail: raldhaheri0001@gmail.com

Received Date: *01 Jul 2022*
Accepted Date: *26 Jul 2022*
Published Date: *01 Aug 2022*

Citation:
Alshareef T, Alfriedy R, Aldhaheri R, Arab N, Badahdah F. A Giant Squamous Cell Carcinoma in a Patient with Pilonidal Sinus: A Case Report. Ann Clin Case Rep. 2022; 7: 2272.
ISSN: 2474-1655.

Introduction

Pilonidal disease is a very common surgical problem that occurs twice as often in men as in women, usually between the ages of 15 and 30 years [1]. Inflammatory complications of pilonidal disease such as cellulitis, abscess, and recurrent sinus tracts are well known to general surgeons [2]. In rare cases, long-lasting inflammation can lead to malignant transformation of the disease, most commonly to Squamous Cell Carcinoma (SCC) [3-6]. The incidence of carcinoma development in pilonidal disease is less than 0.1% and is associated with a high recurrence rate and poor prognosis [7,8]. Carcinomas may show uncontrollable growth and cause substantial disfigurement. Giant cutaneous SCCs larger than 5 cm in diameter are uncommon and can be very difficult to treat [9,10]. Total surgical resection with free margins is the treatment of choice for SCCs arising from pilonidal disease, and tissue defects can be repaired using rotational flaps or skin grafts [11]. Here, we present a case of a patient who underwent excision of a huge cutaneous tumor arising from pilonidal sinus on the midline of the gluteal region with an optimal clinical result.

Case Presentation

The patient was a 66-year-old male retired truck driver with a 5-year history of a growing cutaneous mass on the midline in his gluteal region who was admitted to the general surgery unit. The mass was a protruding, fungating, ulcerated, firm, cauliflower-like tumor. The lesion measured 6.5 cm × 14.2 cm × 16.8 cm that had developed over a large erythematous skin area (Figure 1). He reported that the tumor developed and reached that size within a 5-year period. However, 10 years ago, he had developed Pilonidal Sinus (PNS), for which he did not seek medical advice. Instead, the PNS was managed by herbal remedies and moxibustion, causing irritation to the area. Thereafter,

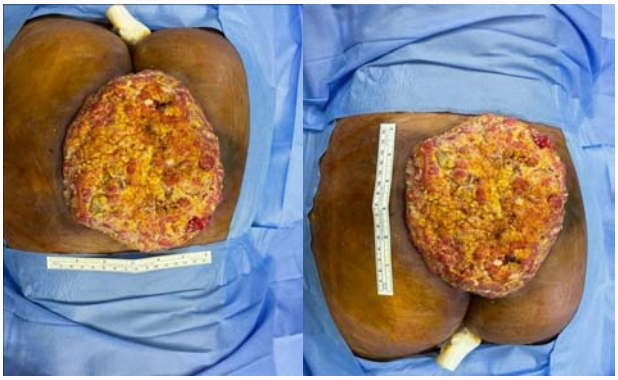

Figure 1: A mass before excision measuring 6.5 cm in width (A) and 16.8 cm in length (B).

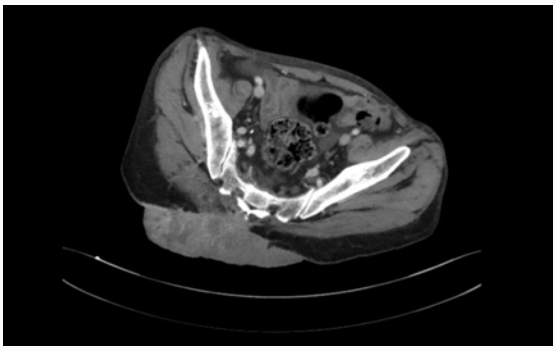

Figure 2: CT scan showing absence of inguinal or presacral lymph node enlargement.

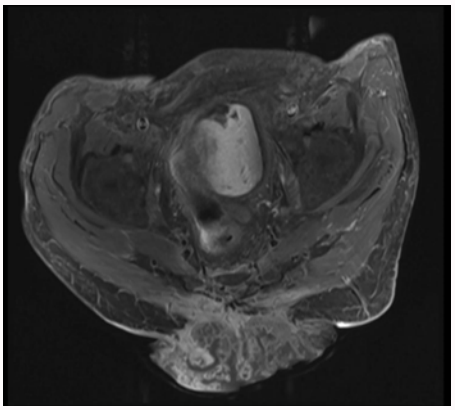

Figure 3: MRI showing the extent of the sinus reaching the gluteal muscle.

the PNS burst open with foul-smelling pus and bloody discharge. The patient recognized that the skin lesion had developed from the PNS but neglected treatment. He finally sought medical advice because of recurrent bleeding, a foul stench emanating from the lesion, and troublesome wound care. He had a history of weight and appetite loss but did not have a history of fever or changes in bowel habits.

Examination

The patient was conscious at the time of examination; his condition was vitally stable and afebrile; oriented. The examination findings were as follows: Temperature, 36.8°C; heart rate, 93 beats per minute; respiratory rate, 20 breaths per minute; blood pressure, 106/60 mmHg; SpO_2, 99%; and weight, 50 kg. A perirectal digital examination showed impacted stool, a normal sphincter tone, and no perirectal bleeding, masses, or ulcer. The patient was admitted for observation and further investigations and administered analgesia, antibiotics, and gastrointestinal and deep vein thrombosis prophylaxes. A punch biopsy was performed for the ulcer margin, the histological examination of which revealed a moderately differentiated, keratinizing SCC of stage T4N0Mx according to American Joint Committee on Cancer staging. Preoperative work-up included a CT scan of the pelvic and abdomen, which showed a posterior superficial cutaneous enhancing mass extending deeply to the gluteal muscle and sacrum with no intraperitoneal extension associated with possible bony erosion but no clear destruction. No lymph node or distant metastases were noted on imaging (Figure 2). No suspicious pulmonary nodules were identified in chest CT. Lumbosacral spin radiography showed mild degenerative changes of the spine, and no other detectable abnormality was identified. MRI of the pelvis indicated a giant exophytic cutaneous gluteal mass with aggressive behavior that showed early infiltration to the adjacent subcutaneous fat, muscles, and bone. No extension to the rectum was observed (Figure 3). A Positron Emission Tomography (PET) scan showed a large hypermetabolic posterior subcutaneous soft tissue mass lesion in the midline of the gluteal region. Hypermetabolic small lesions in the third part of the duodenum may represent small bowel Gastrointestinal Stromal Tumor (GIST), while ectopic pancreatic tissue is another possibility. A small mass lesion with low 18-Fluoro-2-Deoxyglucose (FDG) avidity is likely to be benign in nature. A multidisciplinary team met with an oncologist and a plastic surgeon, and a decision was made to perform total surgical resection of the mass and coverage with a rotational locoregional flap, skin graft, and Vacuum-Assisted Closure (VAC) application.

Hospital course

The patient was malnourished and showed low albumin levels (18 g/L). Therefore, he was started on a high-protein, high-calorie diet. Furthermore, he was losing sleep, had a poor oral intake, and was diagnosed with anxiety disorder during his hospital stay; mirtazapine 7.5 mg PO HS was started. He consented to an operation and after induction of general anesthesia; he was placed in the prone position and underwent preparation and draping under sterile conditions. The tumor was totally excised layer-by-layer till the sacral bone was reached. with 0.3 cm from the deep margin, 1 cm away from the right mucosal margin, 1.5 cm away from the left mucosal margin, 1.5 cm away from the superior mucosal margin, and 2 cm away from the inferior mucosal margin around it (Figure 4) next, freshly frozen sections were sent for histological analysis, and the results were negative (free margin). A histological examination of the surgical

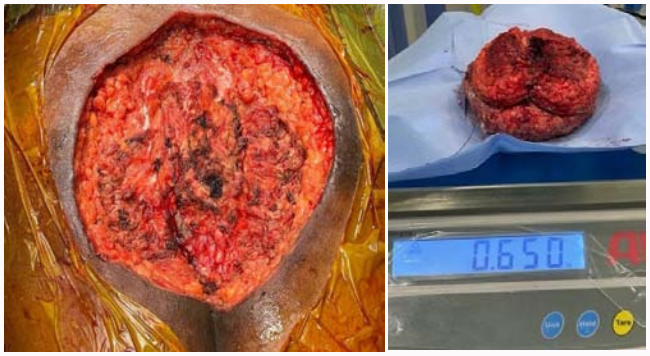

Figure 4: Gluteal area after mass excision (A), weighting 0.65 kg (B).

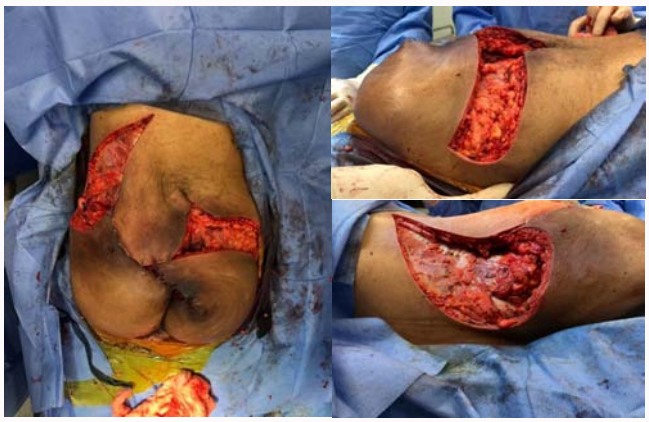

Figure 5: Wound approximation coronal view (A), Right side view (B), Left side view (C).

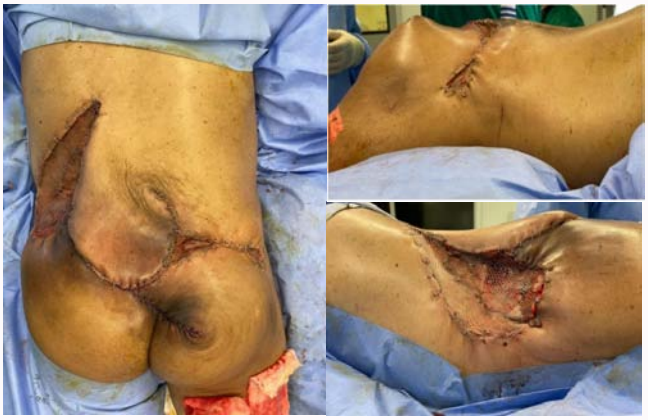

Figure 6: Rotational locoregional flap and skin graft coronal view (A), Right side view (B), Left side view (C).

specimen revealed a keratinizing moderately differentiated infiltrative cutaneous SCC infiltrating deep into underlying subcutaneous tissue by 0.6 cm and a maximum tumor thickness of 5.3 cm. The tumor did not show lymphovascular or perineural invasion. Excision appeared complete in the sections examined. Hemostasis was secured. However, a wide tumoral excision of the surrounding skin could not be approximated (Figure 5), necessitating subsequent flap coverage. Therefore, the plastic surgery team joined the OR for wound closure of the left back and the sacral area. The flap was used for defect coverage; thus, a skin graft taken from the posterior thigh and a temporary vacuum-assisted closure system was placed (Figure 6). The postoperative course was uneventful and the patient's condition improved significantly (Tables 1-3). The wound was taken care of daily, and the patient was discharged home after 1 week on oral amoxicillin-clavulanate. For his malnutrition, he was recommended to consume a high protein and calorie diet with Ensure plus Advance and Resource Fruit Flavored Beverage, and he was also recommended to undergo psychiatric referral regarding his anxiety disorder.

Discussion

Pilonidal sinus disease is a long-standing inflammatory disease that often arises from the hair follicles of the natal cleft of the sacrococcygeal area [12]. Pilonidal sinus recurrence is a matter of concern to both patients and healthcare providers [13]. Its risk factors include obesity, sedentary lifestyle, family history, hirsute body habits, and trauma or irritation to the gluteal cleft skin [11]. SCCs arising in a pilonidal sinus are extremely rare complications of a common disease. SCCs have been shown to develop more frequently from chronic precursor lesions, including chronic ulcers, sinus tracts, or burns [14]. We observed this phenomenon in our patient, with the patient's son reporting that the mass started growing in the patient's buttock and that the patient noticed it arising from the PNS over five years of multiple trials with herbal remedies and moxibustion. Thus, the duration of the disease is a significant factor influencing the malignant transformation of a pilonidal sinus [2]. The present case is extraordinary in many respects, considering the site of the tumor and the fact that the buttock is not a sun-exposed site. With regard to the molecular basis, the pathogenesis was considered to involve the release of Reactive Oxygen and Nitrogen Species (RONs) produced by inflammatory cells at sites of infection inducing DNA damage. Moreover, RONs can also amplify inflammatory responses, leading to increased DNA damage that predisposes patients to malignancy. Immunosuppression and human papilloma virus infection may also be predisposing factors inducing this transformation [15-17]. Almost all tumors arising from pilonidal sinuses are SCCs (90%). Uncommon subtypes such as basal cell carcinomas, adenocarcinoma, and verrucous carcinomas have also been reported. Preoperative evaluations in such cases should include historical examination of an excised or untreated pilonidal disease along with clinical inspection and rectal and sigmoidoscopic examinations [18,19]. In the present case, malignant transformation was suspected due to the recurrence, chronicity, and delayed healing. However, the diagnosis was confirmed by biopsy. Since carcinoma tends to be deeply invasive, preoperative assessment of the degree of invasion by physical examination and endoscopic examination complemented by CT scans or MRI for the sacral region are crucial. In our case, the mass extended deeply to the gluteal muscle and sacrum with no extension to the rectum. Although distant metastases are rare since most cases tend to be locally extensive, positron emission tomography is a good tool to exclude metastases. CT scans or MRI in addition to careful palpation of inguinal lymph nodes can be used to assess intra-abdominal metastasis, including the spread to the iliac and para-aortic lymph nodes [18,19]. In our case, preoperative MRI and PET scan showed negative findings for metastasis. According to a literature review, en bloc resection including at least a wide margin of skin, presacral fascia, subcutaneous fat tissue, gluteal muscle, and, if required, bone is the treatment of choice [5]. Prophylactic inguinal node dissection is not recommended. Radiation alone or postoperatively has not been beneficial in trial reviews, although local control has been obtained for moderate periods of time [2]. The decision in our case was to only perform a total surgical resection of the mass. The resulting defect can be repaired by performing primary or delayed reconstruction. Depending on the size of the postoperative defect, skin grafts or local flaps are being used for the reconstruction [15]. In the present case, the plan was to cover the defect with a rotational locoregional flap, skin graft, and VAC application. Recurrence rates can be as high as 40% to 50%, and recurrence usually appears 9 to 16 months after surgery [6]. The 5-year survival rate of patients with SCCs arising from pilonidal sinus disease is almost 55% to 61% [20]. Metastasis appears in 14% of patients and is usually fatal [2]. Considering the high recurrence rate, regular postoperative check-ups are recommended every 3 months during the first 2 years, every 6 months for the next 3 years, and yearly afterwards [21]. These checkups should consist of physical examination along with abdominal CT scans to exclude local recurrence or distant metastasis [22].

Table 1: Labs at admission: (WBC) White Blood Cell, (BUN) Blood Urea Nitrogen, (PT) Prothrombin Time, (INR) International Normalized Ratio, (APTT) Activated Partial Thromboplastin, (Al kph) Alkaline phosphatase.

WBC	Haemoglobin	Platelets	Sodium	Potassium
7.97 × 10^9/L	8.7 g/dL	500 × 10^9/L	135 mmol/L	3.4 mmol/L
Creatinine	BUN	PT	INR	APTT
84 mmol/L	4.1 mmol/L	17.9	1.3	32
Bilirubin total	Alk. Ph	calcium	phosphorus	Chloride
3 µmol/L	116 U/L	3.18 mmol/L	0.91 mmol/L	100 mmol/L

Table 2: Tumour marker (CA 19-9) Cancer Antigen 19-9, (CEA) Carcinoembryonic Antigen, (CA 125) Cancer Antigen 125, (PSA total) Prostate Specific Antigen total, (PSA free) Prostate Specific Antigen free, (PSA ratio) prostate specific antigen ratio.

CA 19-9	CEA	CA 125	PSA total	PSA free	PSA ratio
17 U/mL	2.2 U/mL	22 U/mL	2.640 ng/mL	0.443 ng/mL	16.78

Table 3: Labs at discharge (WBC) White Blood Cell, (BUN) Blood Urea Nitrogen, (PT) Prothrombin Time, (INR) International Normalized Ratio, (APTT) Activated Partial Thromboplastin, (Al kph) Alkaline Phosphatase.

WBC	haemoglobin	Platelets	Sodium	Potassium
8.3 × 10^9/L	7.1 g/dL	435 × 10^9/L	138 mmol/L	3.7 mmol/L
Creatinine	BUN	PT	INR	APTT
72 mmol/L	3.7 mmol/L	18.3	1.4	38
Bilirubin total	Alk. Ph	calcium	phosphorus	Chloride
5 µmol/L	148 U/L	1.81 mmol/L	0.52 mmol/L	107 mmol/L

Conclusion

In conclusion, we report a rare case of massive SCC of the gluteal region at the midline. Localized SCC of the skin can reach an enormous size if neglected. The invasiveness of the tumor depends on the size, anatomical location, and histological subtype. Moreover, massive SCCs are accompanied by the risk of disfigurement, local recurrence, and metastasis. Finally, surgery is the mainstay of treatment even in giant tumors and should be followed by radiotherapy to address the risk of metastatic spread.

References

1. Abboud B, Ingea H. Recurrent squamous-cell carcinoma arising in sacrococcygeal pilonidal sinus tract: Report of a case and review of the literature. Dis Colon Rectum. 1999;42(4):525-8.
2. Michalopoulos N, Sapalidis K, Laskou S, Triantafyllou E, Raptou G, Kesisoglou I. Squamous cell carcinoma arising from chronic sacrococcygeal pilonidal disease: A case report. World J Surg Oncol. 2017;15(1):1-5.
3. de Parades V, Bouchard D, Janier M, Berger A. Pilonidal sinus disease. J Visc Surg. 2013;150(4):237-47.
4. Martino A, De Martino C, Pisapia A, Maharajan G, Evangelista M. Squamous-cell carcinoma arising in a pilonidal sinus: Case report and review of the literature. BMC Geriatr. 2011;11(1):A28.
5. Mayol Oltra A, Boldó Roda E, Lozoya Albacar R, Morillo Macias V, Nobleja Quiles N. Squamous cell carcinoma over pilonidal chronic disease. A new therapeutic approach. Int J Surg Case Rep. 2020;70:172-7.
6. Pilipshen SJ, Gray G, Goldsmith E, Dineen P. Carcinoma arising in pilonidal sinuses. Ann Surg. 1981;193(4):506-12.
7. Konrad Wroński KX. A rare case of squamous cell carcinoma arising from chronic sacrococcygeal pilonidal disease. Ann Ital Chir. 2019;8:S2239253X19030202.
8. Eryılmaz R, Bilecik T, Okan I, Ozkan OV, Coşkun A, Sahin M. Recurrent squamous cell carcinoma arising in a neglected pilonidal sinus: Report of a case and literature review. Int J Clin Exp Med. 2014;7(2):446-50.
9. Yahyaoui M, Abbassi N, Derfoufi A. Giant squamous cell carcinomas of the shoulder. Pan Afr Med J. 2020;36(215):1-6.
10. Ricci F, Paradisi A, Fossati B. Giant neglected squamous cell carcinoma of the skin. Dermatol Ther. 2015;28:230-4.
11. Pyon RE, Mazumder A, Almajali F, Wong S. Chronic pilonidal cyst with malignant transformation: A case report and literature review. Cureus. 2022;14(3):e23248.
12. Varnalidis I, Ioannidis O, Paraskevas G. Pilonidal sinus: A comparative study of treatment methods. J Med Life. 2014;7(1):27-30.
13. Albabtain IT, Alkhaldi A, Aldosari L, Alsaadon L. Pilonidal sinus disease recurrence at a tertiary care center in Riyadh. Ann Saudi Med. 2021;41(3):179-85.
14. Boukamp P. Non-melanoma skin cancer: What drives tumor development and progression? Carcinogenesis. 2005;26(10):1657-67.
15. de Bree E, Zoetmulder FA, Christodoulakis M, Aleman BM, Tsiftsis DD. Treatment of malignancy arising in pilonidal disease. Ann Surg Oncol. 2001;8(1):60-64.
16. Kidane D, Chae WJ, Czochor J. Interplay between DNA repair and inflammation, and the link to cancer. Crit Rev Biochem Mol Biol. 2014;49(2):116-39.
17. Rassi Z El, Aoun C, Maalouf H. Management of squamous cell carcinoma in a chronic sacrococcygeal pilonidal cyst: A case report. Surg Case Rep. 2020:1-4.
18. Matsushita S, Ohtake N, Mochitomi Y, Fukumitsu K, Nishi M, Kanzaki T. A case of squamous cell carcinoma arising in a pilonidal sinus. J Dermatol. 2002;29(11):757-8.
19. Pekmezci S, Hiz M, Saribeyoglu K, Akbilen D, Kapan M, Nasirov C, et al. Malignant degeneration: An unusual complication of pilonidal sinus disease. Eur J Surg. 2001;167:475-7.
20. Nunes LF, Castro Neto AKP, Vasconcelos RAT, Cajaraville F, Castilho J, Rezende JFN, et al. Carcinomatous degeneration of pilonidal cyst with sacrum destruction and invasion of the rectum. An Bras Dermatol. 2013;88(6):S59–62.

21. Erkent M, Şahiner İT, Bala M. Comparison of primary midline closure, Limberg flap, and Karydakis flap techniques in Pilonidal sinus surgery. Med Sci Monit. 2018;24:8959-63.

22. Parpoudi SN, Kyziridis DS, Patridas DCh. Is histological examination necessary when excising a pilonidal cyst? Am J Case Rep. 2015;16:164-8.