

CARCINOMA ARISING IN PILONIDAL DISEASE – REPORT OF TWO CASES

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Summary. Squamous cell skin carcinoma is a rare complication of pilonidal disease. The signs of malignant change are induration, rapid growth and sharp pain in ulcerated tumor. Men are mostly involved. Histology defined highly differentiated squamous carcinoma in one and a bad differentiation in the other. Carcinoma in pilonidal disease mainly arises after 15 year disease history. Two men were operated on for this carcinoma at Surgical clinic of Nis. After biopsy, carcinoma was operated radically (large excision) and margins needed to be clean. Immediate reconstruction was performed with skin grafts and musculocutaneous gluteus maximus peddiced flap. Postoperative radiation therapy was performed. One patient is disease free after 3 years, but other died 9 months after surgery. The primary prevention from carcinoma is surgery for early pilonidal disease. Large surgical resection and clean margins could obtain satisfactory results.

Key words: Pilonidal disease, skin carcinoma, surgery

Introduction

The most common complications of pilonidal disease are cellulitis, abscess or recedivant infection and fistula forming. Malignant degeneration and carcinoma emerged on the terrain of fistulised pilonidal disease are extremely rare complications and this condition is described within the group of Marjolin cancer. These carcinomas appear on the region of chronic irritation or on scars (burn scars which epithelised spontaneously, or osteomyelitic fistula). Carcinoma can also be seen on a place of a long-term infection.

The treatment of carcinoma requires large surgical excision with margins at least 3 cm into the healthy tissue and immediate reconstruction performed with skin grafts or rotational flap.

If there are positive lymph nodes in inguinal region dissection is being done.

Objective

We present 2 cases of patients treated for squamous cell skin carcinoma as a complication of pilonidal disease at Surgical clinic of Nis in the period of 5 years.

Case I

A 57-year-old male was admitted to hospital in May 2002, with large, ulcerated skin tumor in sacrococcygeal

region. Since the age of 40, when he had been diagnosed pilonidal disease, he had had 12 incisions and abscess evacuation, and each time he refused surgical treatment. In the past year ulceration had appeared, and occasionally it bled both spontaneously and on pressure, also there was an unpleasant odorous secretion from it. Sharp pain occurred two months ago and now it could not be controlled by taking oral analgetics.

Clinical examination showed ulcerated tumor induration with 12cm in diameter which bleeds. General condition of the patient is satisfactory and he has no other diseases. Lymph nodes in inguinal regions are not enlarged. By digitorectal examination no induration towards rectum was found. Laboratory findings include leucocyte count of 13,000. Radiography of lumbosacral spine and pelvis is normal. As infection prophylaxis cephalosporins were given for three days before the operation. Large tumor excision and immediate reconstruction using skin grafts and local skin flap was performed on May 27th 2002.

Postoperative period was regular. The skin graft was primary accepted in 90% of and the remaining defect epithelised spontaneously through regular bending (Fig. 1, 2).

Histology defined highly differentiated squamous cell skin carcinoma. Resection margins were clear (no tumor was found).

Postoperative radiation therapy was performed. Patient was regularly controlled in the next three years and

no signs of disease progression were found. He died of myocardial infarctus in may 2005.



Fig. 1. Case 1-Preoperative view - Pilonidal carcinoma



Fig. 2. Case 1- Postoperative result

Case II

A 49-year-old male was admitted to hospital in October 2005 with a complaint of the ulcerated skin tumor in sacrococcygeal region. He had had pilonidal disease ever since he was 28, and a vast number of pilonidal abscess incisions, but five years ago fistula occurred which he drained by himself. He had been refusing operation. But now he had sharp pain and significant dis-

comfort while sitting or lying. He denied any previous disease or allergy. Clinically, there was a tumor in sacrococcygeal region with marked induration measuring about 8 cm in diameter, and an ulcer in the middle covered with mixture of necrotic detritus and blood (Fig.3). Tumor was fixated by the sacrum. Lymph nodes in both inguinal regions were not enlarged. Digitorectal examination showed extraluminal compression on the rectum from the back side. Laboratory data showed leucocytosis. After tumor biopsy, pathologic studies revealed squamous cell skin carcinoma. After CT scan of pelvis and abdomen, coccygeal destruction and sacrum infiltration in distal fifth of the bone were found (Fig. 4).



Fig. 3. Case 2- Intraoperative view - tumour excision



Fig. 4. Case 2- CT scan implies sacral destruction

Preoperatively the patient was treated with antibiotics and local wound dressing for three days. Then he underwent a surgical procedure. After tumor excision, parts of anal canal were excised and a tumor mass from small pelvis was extracted (Fig. 5). Clinically, tumor was excised in whole all the way to the healthy tissue by excision sacrovesical septums. Then a resection of a distal part of the sacrum 2,5 cm from the edge of the

bone defect was performed. Postoperative defect was closed with musculocutaneous gluteus maximus peddled flap on the right, and fasciocutaneous flap on the left side (Fig. 6). Pathology revealed badly differentiated squamous cell skin carcinoma. All margins were free of disease. Secondary defect was covered by skin grafts. At the end of procedure protective sigmoidostomia was made (Fig. 7).

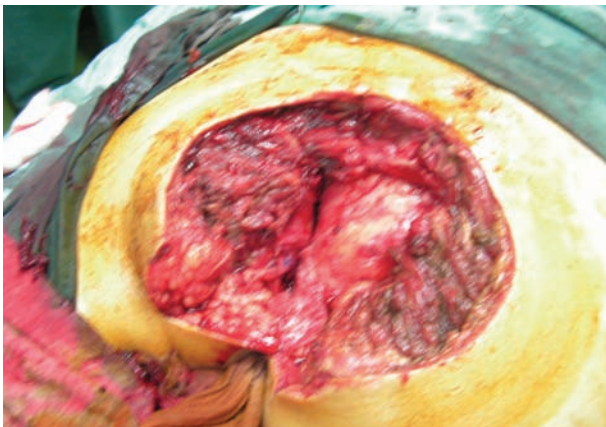


Fig. 5. Case 2- Intraoperative view- tissue defect



Fig. 6. Case 2- Defect is reconstructed by musculocutaneous gluteus maximus flap and 1 skin flap

The postoperative course was regular, with no abdomino-perineal complications. The wound healed secondary, because of graft infection in secondary defect. That wound was healed by secondary intention. A month after, rectal fistula appeared. The patient underwent postoperative radiotherapy. Control CT scan two months after the operation showed no signs of local recurrence. Six months after, patient was readmitted to hospital because of the recurring disease locally described as 6cm in diameter infiltrating rectum and bladder. It was established by the oncological team that the tumor was inoperable in this phase, so the patient was treated with nonspecific adjuvant therapy. Unfortunately, the patient died 9 months after the surgical procedure.



Fig. 7. Case 2- Definitive sigmoidostomy

Discussion

The first squamous cell skin carcinoma arising in pilonidal sinus was described in 1900 (1). The frequency of the carcinoma is very low and in the whole medical literature less than 100 patients are reported. The incidence of carcinoma arising in pilonidal disease is less than 0,1%, on anal fissure is 0,1% and with osteomyelitic fistula is 0,2% (2). Available literature stated out that the carcinoma is more often in males, the average age at diagnosis is 52 years and the mean duration of pilonidal disease before malignant alteration is 20 years (3). Pilonidal carcinoma is described in higher rate in immunocompromised patients (receiving immunosuppression) (4). Our cases were 49 and 57 years old, which correlates with literature data.

Pilonidal carcinoma is diagnosed by clinical examination as a growing, ulcerated mass with indurated edges, which is very painful and often bleeds. The connection between carcinoma arising and pilonidal disease is obvious from the anamnesis, and diagnosis is confirmed by preoperative biopsy specimen analysis. The most important ethiological factor is chronic infection and skin irritation as in osteomyelitic fistula (5). Both patients had pilonidal disease for many years and refused surgical treatment.

Preoperative diagnosis standards are standard radiography of pelvis and CT scan of small pelvis and sacral region. Rectal examination is obligatory. Antibiotic therapy was preoperatively given (6, 7). Preoperative protocol was carried out as a routine and both patients were treated with prophylactic antibiotic therapy using cephalosporins.

Surgical procedure includes tumor excision to the healthy tissue. Clinically healthy tissue margin excision involving skin, subcutaneous tissue and gluteal muscle must be at least 3 cm. Sacral fascia is needed to be removed, as well as coccygeal excision and decortication or resection of the sacrum is needed to be performed if there is bone infiltration on CT scan (8). The patient who had infiltration within the bone structures underwent resection of the distal fifth of the sacrum.

Elective inguinal node dissections have not been recommended. Groin dissections are performed in cases of both clinically and positive lymph nodes in that area on sonography. In the reported cases there was not any lymph node enlargement found in the inguinal region.

Defect reconstruction could be performed primary 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 (8). In one case postoperative defect was closed with skin grafts, and in other with musculocutaneous gluteus maximus pedicled flap, local skin flap and skin graft.

After surgery, the radiation therapy is recommended (8). Both our patients were treated with radiation therapy. Postoperative follow-up protocol includes regular check-ups by the oncology expert committee: every three months in the first two years, then every 6 months till the end of the fifth year and once a year after that.

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KARCINOM NASTAO IZ PILONIDALNOG SINUSA – PRIKAZ DVA SLUČAJA

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Kratak sadržaj: Karcinom na pilonidalnom sinusu je retka komplikacija bolesti. Sumnju hirurga treba da pobudi induracija i rast ulcerisanog tumora kao i pojava jakih bolova. Češće se javlja kod muškaraca. Patohistološki je dobro diferentovan planocelularni karcinom kod jednog, a kod drugog loše diferentovan. Ovaj tumor nastaje kod pacijenata sa dugogodišnjom anamnezom (preko 15 godina). Dva pacijenta sa pilonidalnim karcinomom su operisana u Hirurškoj klinici u Nišu. Hirurško lečenje pacijenta sa karcinomom na pilonidalnom sinusu zaheva široku radikalnu hiruršku eksciziju uz kontrolu resekcioničkih ivica i primarnu rekonstrukciju defekta. Rekonstrukcija defekta je rađena

transplantatima kože i režnjevima. Postoperativno je sprovedena zračna terapija. Jedan bolesnik posle 3 godine nema znakove za recidiv niti metastaze, a drugi bolesnik je umro nakon 9 meseci od operacije. Najbolja prevencija karcinoma nastalog iz pilonidalnog sinusa je hirurško lečenje pilonidalne bolesti u ranoj fazi. Radikalna hirurška ekscizija sa kontrolom ivica resekcije može obezbediti uspeh u lečenju.

Ključne reči: *pilonidalna bolest, karcinom kože, hirurgija*