



Squamous cell carcinoma and pilonidal cyst disease

Case report



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Squamous cell carcinoma and pilonidal cyst disease. Case report

AIM: Squamous cell carcinoma developed on a chronic pilonidal cyst.

CASE REPORT: Authors describe the case of a squamous cell carcinoma developed on a chronic pilonidal cyst in a 63-years-old patient with a 43 years history of recurrent pilonidal sinus disease.

RESULTS: The patient underwent incisional biopsy, staging with total body CT and, finally, radical surgery. After 30 months there were no evidence of recurrence.

DISCUSSION: Pilonidal sinus disease is a common disease that affects especially male subjects, obese and with excess of body hair. The complications that arise most frequently are cellulitis, abscess formation and developments of recurrences. Malignant transformation appears rather rare and is reported in the literature with a percentage that goes from 0.02% to 0.1%.

CONCLUSIONS: Authors recommend accurate inspection of the pilonidal area in all chronic and longstanding inflammatory lesions and possibly practice incisional biopsies to exclude malignant degeneration.

KEY WORDS: Pilonidal sinus, Squamous cell carcinoma, Skin flaps

Introduction

Malignant transformation on sinus pilonidalis is a very rare occurrence. The most common type is Squamous cell carcinoma in more than 80% of cases, followed by Basal cell carcinoma, less than 10%, and even more rarely mixed or unclassified forms of adenocarcinoma¹. In the

literature are reported less than 70 cases of squamous cell carcinoma related to pilonidal cysts². All these patients have a long history, more than 10 years, of abscess and between the pilonidal sinus' and the carcinoma's diagnosis often occur an interval of about 23 years. The therapeutic choice consists in a surgical excision of the tumor tissue, associated, according to some authors, with adjuvant chemotherapy and radiotherapy³⁻⁴. We describe the case of a squamous cell carcinoma in a patient who has had a pilonidal cyst for 44 years.

Case report

A.E., 63-years-old, male. He tells about the common childhood rashes, a history of hypertension in treatment,

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GERD (*Gastro-Esophageal Reflux Disease*) in treatment. He undergoes in 1969 a first surgery for a sacrococcygeal fistula. The patient tells about repeated episodes of sacrococcygeal fistula's abscess, treated with empirical antibiotic therapy. He comes to our observation after one of these episodes, with suppuration and numerous fistulous tracts that affect, bilaterally, the perineal area (Fig. 1). We carry out drainage and a further antibiotic therapy, but because of the failure of this strategy, in April 2012, the patient undergoes an incisional biopsy. An elliptic incision that includes the button-like ostium of one of the sinus tracts is performed. A massive inflammatory process with limited purulent collections is found

and the excision is extended to reach tissues that are acceptably free from macroscopic disease. The wound is cleaned with iodine solution and filled up with gauze in order to let it heal by secondary intention. Histological examination of the surgical sample reveals a diffuse infiltration of well and moderately differentiated squamous cell carcinoma, which affects the deep edge of the sample, too. We programme staging with TC, to value the local extent of the disease and the possibility of intra-abdominal metastasis, and adjuvant radiotherapy. Following up the disappointing recovery, is planned a further surgery in October 2012. It is performed an excision of a lozenge of skin of about 11,5 x 6,4 x 6 cm, with abundant adipose tissue, centred on an area with crater-like aspect, including a portion of the coccyx near the deep margin, trying to preserve the integrity of the anal sphincter and, at the same time, to obtain margins free from disease (Figs. 2, 3, 4). The wound is closed by the use of flaps (Figs 5, 6, 7).



Fig. 1: Clinical presentation

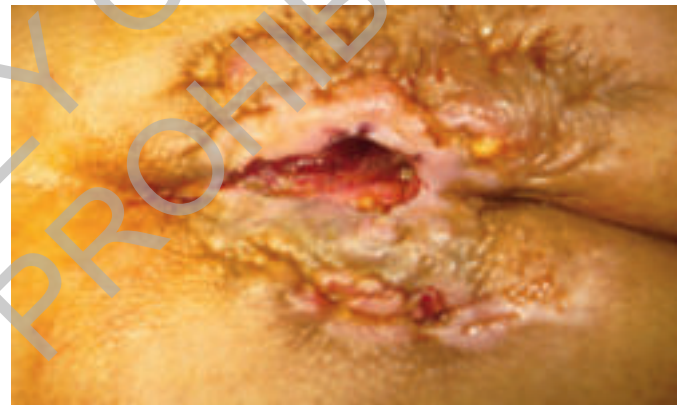


Fig. 3: Closeup of skin lesion



Fig. 2: Skin lesion after first treatment

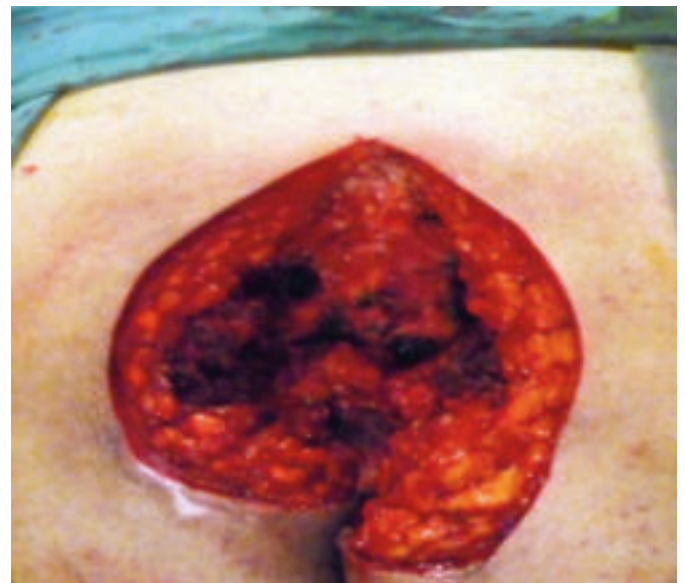


Fig. 4: View after lesion removal



Fig. 5: Preparation of skin flaps 1



Fig. 7: Wound closure

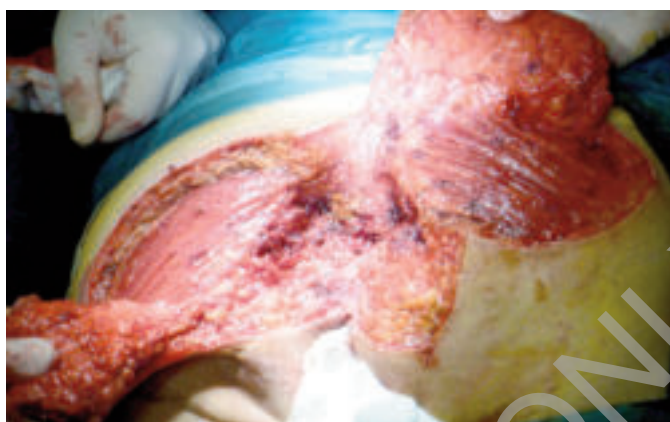


Fig. 6: Preparation of skin flaps 2



Fig. 8: Results 30 months after surgery

Results

Histological studies describe an infiltrating well/moderately differentiated squamous cell carcinoma with excision margins free from tumour. In agreement with the oncologist, the radiotherapy was considered not necessary. After 30 months there were no evidence of recurrence (Fig. 8).

Discussion

Pilonidal sinus disease is a common disease that affects especially male subjects, obese, with occupations that require long-sitting positions, excess of body hair, lack of body hygiene and profuse sweating⁵. The complications that arise most frequently are cellulitis, abscess formation and developments of recurrences. The rare complications include sacral osteomyelitis and meningitis. Malignant transformation appear rather rare and is reported in the literature with a percentage ranging from 0.02% to 0.1%². The average age at diagnosis is about 50 years old, with an history of pilonidal sinus disease

lasting about 2 decades. The continuous process of tissue damage and repair related to the chronic inflammation, during which the mechanism of DNA repair is compromised, seems play a role in development of the squamous cell carcinoma⁶. As reported by many authors, pilonidal carcinoma has a characteristic aspect with a central ulcer with hard, brittle and irregular margins, and in many cases in continuity with the cyst⁴⁻⁷. The clinical case that we have presented did not clearly show these characteristics so that it was difficult to diagnose a carcinoma before the surgery. During the physical examination it is necessary the careful examination of perineum, anus and both inguinal regions. It is also recommended the execution of a preoperatively colonoscopy and a pelvic CT scan in order to value the local and remote spread of the disease¹. The main treatment option is represented by wide excision of the lesion, and this must, eventually, be followed by adjuvant radiotherapy in those cases where the extent of neoplastic infiltration does not provide sufficient guarantees of surgical radicality. The clinical behaviour of the squamous cell carcinoma is generally aggressive with percentages of recurrence between 34 and 50%⁴⁻³⁻⁸.

Conclusions

In chronic forms of Pilonidal cyst can happen a malignant degeneration following a long period of inflammation of the tissues affected by the disease, though it is a rare complication and less than 70 cases are reported in literature². Local recurrences are frequent and a wide excision followed by an extension in case of relapse or not healing may improve the prognosis *quod vitam*. Various authors have used chemotherapy and radiotherapy before and after the operations but because of the small number of cases reported in literature it is difficult to identify the most effective therapeutic approach and to evaluate the long-term effectiveness of the different treatments.

Summary

The squamous cell carcinoma arising out of pilonidal sinus disease is a rather rare event that occurs in case of long-course pilonidal cyst disease. It's characterised by a slow growth but a high invasiveness. The authors report a case of a 63-years-old patient with decennial pilonidal sinus disease and recurrent abscess formations. The patient underwent to two surgical operations and the second one included a very wide resection and reconstruction by the use of flaps. After 30 months neither complications nor local recurrence were observed.

Riassunto

Il carcinoma a cellule squamose insorgente su malattia del seno pilonidale è una patologia abbastanza rara che sopraggiunge in presenza di malattia con decorso decennale. È caratterizzato da una crescita lenta ma da un'elevata invasività locale. Gli autori riportano il caso di un paziente di 63 anni con storia pluridecennale di malattia del seno pilonidale con ascessualizzazioni ricorrenti trattato chirurgicamente con resezione ampia e ricostruzione mediante uso di lembi. A distanza di 30 mesi non sono state osservate complicanze o recidive locali.

References

1. Alarcón Del Agua, Carlos Bernardos-García C, Bustos-Jiménez M, et al.: *Malignant degeneration in pilonidal disease*. Cir, 2011; 79:346-50.
2. Tirone A, Gaggelli I, Francioli N et al.: *Malignant degeneration of chronic pilonidal cyst. Case report*. Ann Ital Chir, 2009; 80(5):407-09.
3. Pilipshen SJ, Gray G, Goldsmith E, et al.: *Carcinoma arising in pilonidal sinuses*. Ann Surg, 1981; 193:507-12.
4. De Martino C, Martino A, Cuccuru A, Pisapia A, Fatigati G: *Squamous-cell carcinoma and pilonidal sinus disease. Case report and review of literature*. Ann Ital Chir, 2011; 82(6):511-14.
5. Fasching Mc, Meland Nb, Woods Je, Wolff Bg: *Recurrent squamous-cell carcinoma arising in pilonidal sinus tract. Multiple flap reconstructions. Report of a case*. Dis Colon Rectum, 1989; 32(2):153-58.
6. Trent TJ, Kirsner RS: *Wounds and malignancy*. Adv Skin Wound Care, 2003; 16:31-4.
7. Davis Ka, Mock Cn, Versaci A, Lentricchia P: *Malignant degeneration of pilonidal cysts*. Am Surg, 1994; 60(3):200-04.
8. Harlak A, Mentes O, Kilic S, Duman K, Yilmaz F: *Sacroccygeal pilonidal disease: Analysis of previously proposed risk factors*. Clinics, 2010; 65(2):125-31.