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SHORT REPORT

Squamous cell carcinoma associated anal fistulas in Crohn's disease unique case report with literature review

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Abstract

Squamous cell carcinoma arising from perineal fistula in patients with Crohn's disease (CD) is a rare entity, with few reported cases in the literature making its clinical characterization troublesome. The outcome is poor following operative treatment as the malignancy of chronic perineal fistula in Crohn's disease is usually overlooked and tardily diagnosed. We present a unique case of a 47-year-old man with a 20-year history of chronic perineal fistula that was diagnosed with extensive perineal squamous cell carcinoma extending down to the right thigh. Computer tomography showed locally advanced disease with inguinal and lung metastasis. Treatment was consisted of diverting colostomy and palliative care. The patient died 3 months later. The clinical data and clinicopathological features of reported cases in the literature were reviewed.

1. Introduction

Anorectal fistulas are common manifestations of Crohn's disease. The development of carcinomas in chronic anorectal fistulas of CD is considered rare and a separate entity from the increased risk of colonic adenocarcinomas in patients with CD.[1] When compared to the overall incidence of colon cancer in CD, the incidence of cancer in perineal fistulas appears to be quite small. Ky et al. following more than 1000 patients with long-standing CD complicated by anorectal fistulas over a period of 14 years estimates the incidence to be about 0.7% of CD patients.[2] The malignancies developing from anorectal fistula in CD are either squamous cell carcinomas or mucinous adenocarcinomas. Most reports consist of single cases or

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small series and both adenocarcinoma and squamous cell carcinomas are frequently reported together making the thorough description of each entity difficult. Approximately 20 cases of squamous cell carcinoma arising in anal fistula in Crohn's disease were reported in English literature from 1980 to April 2012.[2–15] Most of those cases were reported from Northern Europe, North America and East Asia. As far as we know, there are no reports from Africa. In University Hospital of Fes, Morocco since 1991, about 340 patients were treated for Crohn's disease. We herein present a unique report of extended squamous cell carcinoma developed from long standing complex anorectal fistula of Crohn's disease along with review of reported cases in literature.

2. Case report

A 47-year-old man with a 20-year history of chronic perianal fistula. The patient resisted surgical treatment until he presented with multiples perianal abscesses. On clinical examination, he was found to have a right hemi circumferential stricture of the anus with a perianal abscesses associated with multiple discharging fistulas. Multiples drainages incisions were performed with seton placement in fistulas tracts. Biopsy of perianal fistula tracts revealed a signs of Crohn's disease. Further investigation including colonoscopy, gastroduodenal endoscopy with multiples biopsies, small bowel magnetic resonance imaging (MRI) did not show any evidence of inflammation or CD in gastrointestinal tract. Perineal MRI showed multiples Transphincteric and suprasphincteric fistula tracts. The patient's Crohn's disease was confined to the perineal area and was treated with 5-ASA, ciprofloxacin and metronidazole with a moderate remission. Azathioprine and Infliximab could not be afforded by the patient because of its high cost. Six months later the patient presented with a new onset of discharging multiple fistula tracts on the area around the left buttock extending down to the left thigh and up to the presacral area. On examination we have noted an extensive perineal fistula opening around the left buttock. The infection extended to the left thigh and presacral area which were both inflamed and discharging pus. Treatment options were discussed with the patient and he declined to have an abdominoperineal resection. After obtaining informed consent, emergency drainage incisions and debridement were performed simultaneously followed by a diverting-loop colostomy. During the next 2 months and despite of fecal diversion, continuous antibiotic treatment along with sulfasalazine, the patient was getting worse with increasing pelvic pain and discharging fistulas. On physical examination the perineal and thigh wounds did not heal and continued to drain permanently, showing an ulcer with budding characteristics (Fig. 1). Multiple Biopsies from the borders of the wounds showed squamous cell carcinoma. A computed tomography (CT) scan of the abdomen and pelvis showed an advanced locoregional disease involving the sacrum, the left iliac bone laterally with inguinal and lung metastaic lymph nodes. MRI of the pelvis confirm the CT scan finding (Fig. 2). Given the findings of squamous cell carcinoma with extensive locoregional and metastatic disease. The tumor was judged unresectable. The patient underwent palliative care. He died 3 months later.

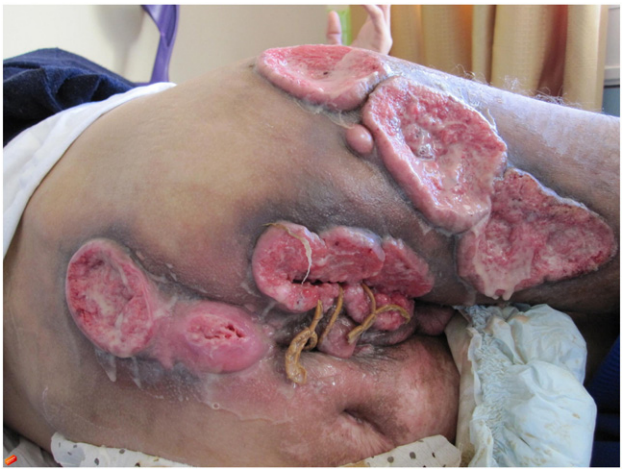

Figure 1 Extensive perianal Crohn's fistula with ulcero budding characteristics.

3. Discussion

Perianal fistulas are found in 20% to 25% of patients with Crohn's disease limited to the ileum and in 60% when the rectum is involved.[16] Patients with perianal Crohn's disease have a 0.7% incidence of carcinoma.[17] Since Peterson was reported the first cases of SCC arising from CD perineal fistula in 1983, we could find 20 other reports in English literature.[2–15] Most reports consist of single cases or small series and both adenocarcinoma and squamous cell carcinoma are frequently reported together making the thorough description of each entity difficult. To our knowledge there are no population-based studies targeting SCC arising from perineal fistula in patients with CD which makes the

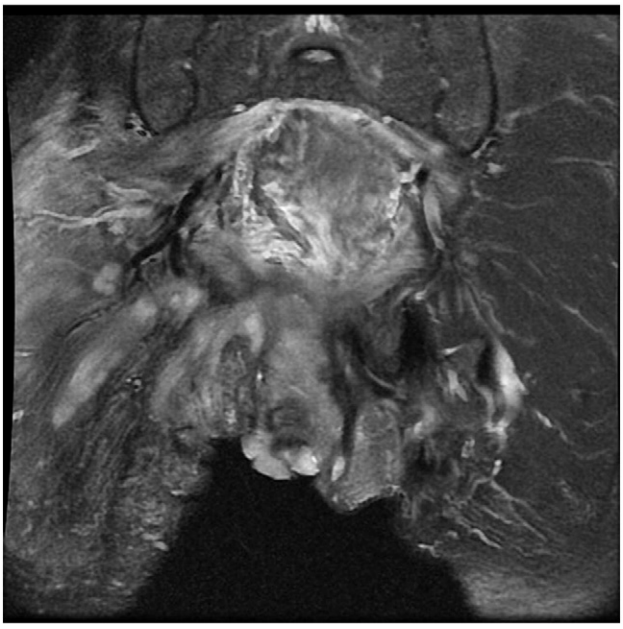

Figure 2 MRI of the pelvis T2 showing advanced locoregional disease.