Neoplastic transformation in longstanding fistula-in-ano

G.H. Welch and I.G. Finlay

University Department of Surgery, Royal Infirmary, Glasgow, G31 2ER, UK.

Summary: A case in which an infiltrating mucinous carcinoma developed within a suprasphincteric fistula-in-ano is presented. The diagnosis was suspected on biopsy and confirmed by repeat biopsy. The clinical and histological features of this case establish with certainty that the carcinoma arose within the fistula and was not a secondary manifestation of the tumour. It is suggested that this rare complication of chronic fistula-in-ano may be prevented by prompt expert management of complex primary fistula.

Introduction

Carcinoma of the rectum may co-exist with, or may present as, a fistula-in-ano. The development of a carcinoma in a longstanding fistula-in-ano, however, is rare.¹ We report a patient with recurrent perianal sepsis and a suprasphincteric fistula-in-ano of 14 years duration, in which a mucinous carcinoma developed.

Case report

A 56 year old man presented with an indurated, scarred perineum with a fluctuant ischio-rectal abscess, discharging via a 5 mm sinus.

Fourteen years previously he had attended another hospital where extensive drainage and debridement had been carried out for an ischio-rectal abscess complicated by necrotizing fasciitis extending onto the lower abdominal wall. Over the ensuing years he had had numerous perianal and ischio-rectal abscesses drained. A high fistula had been demonstrated and a laparotomy and loop colostomy performed. However, at the patient's request the colostomy had been closed prematurely and the patient had been lost to follow up.

Once again the ischio-rectal abscess was drained as an emergency procedure. Subsequently an examination under anaesthesia and fistulagram were carried out which demonstrated the presence of a high suprasphincteric fistula-in-ano (Figure 1). Although the mucosa appeared macroscopically normal, biopsies were taken to exclude the presence of inflammatory bowel disease. The biopsies were reported as showing cellular atypia, but there were no features of inflammatory bowel disease. At the pathologist's request biopsies were repeated from the rectal mucosa and from within the tract. Histological examination

revealed a mucinous adenocarcinoma within muscle. An abdomino-perineal excision of the rectum was carried out.

Gross examination of the specimen revealed diffuse thickening of the rectal wall 6 cm from the skin edge. There was a fistula tract arising from the skin and opening into the rectum 1 cm above the dentate line, but no evidence of mucosal tumour. Histological examination revealed a moderately well differentiated mucinous adenocarcinoma present within muscle and fibrous tissue of the sinus tract. There was no microscopic evidence of tumour within the mucosa or at the skin margin.

Discussion

The occurrence of a carcinoma in association with a fistula is probably due to chronic inflammation, although the rarity of the condition precludes any definite assumption in regard to the aetiological relationship of the fistula and carcinoma. A rectal carcinoma may present as a fistula and it may be difficult to determine whether the tumour is a complication of a longstanding perianal fistula or whether the perianal fistula is merely a manifestation of the malignancy itself.

Rossner² established three essential criteria to confirm that malignant transformation has occurred within a fistula. He stated firstly that the fistula must be present for 10 years to exclude the possibility that the malignancy predated the fistula; secondly, that there should not be tumour within the mucosa of the rectum or anal canal unless there is definite evidence that this is metastatic tumour; and thirdly that the opening of the fistula within the anal canal or rectum should not contain malignant tissue. In the patient presented, the length of the preceding history, in this

Correspondence: G.H. Welch, F.R.C.S.
Accepted: 16 December 1986

Figure 1 Fistulagram showing high fistula-in-ano (arrowed).

case 14 years, and the histological features fulfil Rossner's criteria.

The difficulty in establishing a diagnosis of carcinoma results mainly from the insidious nature of the tumour and the masking effect of the symptoms of the fistula. Forty-four percent of the previously reported cases are mucous adenocarcinomas of low grade malignancy.³ In this case cellular atypia noted at the initial biopsy focused attention on the possibility of underlying malignancy within the tract. Abdomino-perineal excision of the rectum completely excised the tumour which was less than 1 cm in diameter.

Unfortunately there are insufficient reported cases of this type to determine the prognosis for individual patients. However, as radical excision offers the only prospect for cure, the possibility of malignant change within a longstanding fistula-in-ano should be considered and multiple biopsies, including biopsy of the tract itself, submitted for histological examination. Indeed where chronic sepsis secondary to a complex fistula-in-ano persists for over a decade, excision of the rectum may well be an appropriate course of action. Clearly, prompt and expert management of complex fistula-in-ano will pre-empt this problem.

References

1. Kyzer, S., Bayer, I., Turani, H. & Chaimoff, C. Verrucous squamous carcinoma as a complication of recurrent multiple perianal fistulae. *Colo-proctology* 1985, 7: 104–106.
2. Rossner, C. Relation of fistula-in-ano to cancer of the anal canal. *Trans Am Proc Soc* 1934, 65–70.
3. Kline, R.J., Spencer, R.J. & Harrison, E.G. Carcinoma associated with fistula-in-ano. *Arch Surg* 1964, 89: 989–994.