

## Carcinoma Associated with Anal Fistula

by

SYOEI YOH, MAMORU YAMANE and KOICHI ISHIGAMI

The 2nd Surgical Division of Kyoto University Medical School

(Director : Prof. Dr. CHUJI KIMURA)

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Anal cancer is comparatively rare among rectal cancers, constituting only 3-4% of all rectal cancers including anal cancer.

On the other hand, anal fistula is one of the most common diseases and apt to be left without treatment. Some authors reported that a few malignant diseases developed from the chronic sinuses and fistulous tracts. However, the question of whether or not carcinomatous changes are caused by the prolonged inflammatory reaction in such tracts has not been established so clearly.

In 1940, EWING quoted KRASKE's statement, "there is no satisfactory evidence that cancer develops in tissues altered by hemorrhoids, fistulae or cicatrices." Of course, it is impossible to determine whether a malignant tumor arises in a chronic sinus or a fistulous tract, or a chronic sinus or a fistulous tract arises in a malignant tumor.

Recently numerous authors have concluded that chronic sinuses and fistulae do of themselves sometimes give rise to malignant changes. Among them are ROSSER, MILLER and LYPIN, SKIR and McCUNE.

SKIR suggested that the best criteria for determining a causal relationship is the time factor. He arbitrarily set ten years as the period over which a fistula must be present before neoplastic changes develop in it, to prove an etiologic connection between the tumor and the tract. He concluded that one must think of chronicity of a fistula developing the possibility of malignant change.

In 1952, McINTYRE said that the possibility of carcinoma in an anal fistula should be considered when (1) the fistula has been present for many years, (2) the fistula, although continuing to drain, becomes more painful or more indurated and (3) the drainage becomes more mucinous in character.

We have experienced 10 cases of anal cancer among 188 cases of rectal cancer from 1956 to 1967 in our clinic.

The frequency of anal cancer to the rectal cancer including the anal cancer is 5.3%.

In 1954, GRINNEL reported 49 cases in 1026 cases of anal and rectal cancers, accordant to 4%. In 1964, BACON reported 80 cases of anal cancers in 2160 cases including the anal cancers, the rectal cancers and the colon cancers, accordant to 3.6%.

Indeed the occurrence of carcinoma in association with an anal fistula is rare. McANALLY and DOCKERTY estimated the association of carcinoma with anal fistula to be 0.1% in 1949.

In 1964, KLINE reported 79 cases of carcinoma associated with anal fistula in literature including his own two cases.

It is the purpose of this paper to report two cases of carcinoma associated with anal fistula recently experienced in our clinic.

### CASE REPORTS

Case 1. H. Y. sixty six years old, male. About 48 years ago, he received the incision of the periproctal abscess. Since a few years after that an anal fistula had been present for 45 years infrequently draining a small amount of pus without medical treatment. Since one year before his admission he has been suffered from the anal bleeding and the anal pain during his defecation. Since a half year before the admission the drainage has become mucinous in character. Past history and family history were non-contributory. The general physical examination was negative.

Examination of the perianal region and perineum revealed three external fistulous openings with cicatricial induration at the posterior portion of the perianal region apart 5 cm from the anal verge. Mucinous material mixed with pus was draining from only two fistulous openings. By digital examination he complained of severe pain and the finger could hardly insert through cicatricially narrowed canal. Many inguinal lymph nodes swelled in both side.

Preliminary left side colostomy was performed and biopsy from the three fistulous openings revealed papillary adenocarcinoma (Fig. 1). After the preoperative irradiation of the  $^{60}\text{Co}$  (4600 R.) for 3 weeks, an abdominosacral resection including a clearing of the both inguinal lymph nodes performed May 10, 1967, at which time no evidence of extension of the carcinomatous process could be seen within the abdomen. The postoperative pathologic report was: (1) adenocarcinoma, (2) lymph node metastasis were positive on both side.

The patient made an uneventful recovery but one year later he readmitted for regional recurrence and lung metastasis.

Case 2. Y. S. fifty four years old, male.

About twenty five years ago this patient received an operation for the periproctal abscess. Since then an anal fistula had been present for twenty three years infrequently draining a small amount of pus without medical treatment. About three months before admission this patient developed a painful swelling in the fistulous opening. On account of the severe pain he could not take a supine position, then he visited to our clinic.

Past history revealed he had had lung tuberculosis about fifteen years ago: otherwise it was non-contributory. Family history was also non-contributory. The general physical examination was within normal limit. Examination of the perianal region revealed two

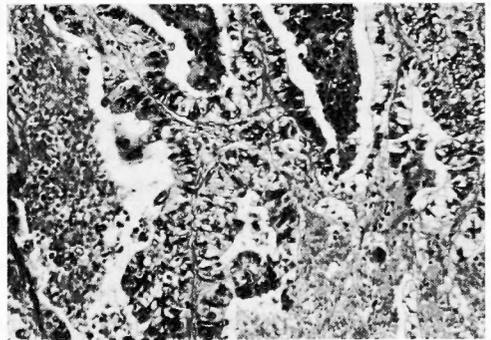


Fig. 1. Histological findings of resected tumor in case 1. Papillary adenocarcinoma (H. E.-staining).

external fistulous openings, one was in right side buttock with a cicatricial deformity and another was in left side buttock associated with a purple red coloured indurated mass 5 cm in diameter and tenderness. The latter fistulous opening was able to be probed 5 cm in length. Digital examination also revealed an indurated mass 5 to 6 cm in diameter, but the mucous membrane overlying this mass was normal.

Sigmoidoscopy was normal. A culture of the fistulous tract was negative for acid-fast bacillus.

A biopsy of the fistulous tract revealed non-specific granulation, so incision to the swelled mass and fistelectomy was performed. The mass was tumorlike in appearance and not pustulous, then a larger section was excised from the mass for the histological examination, which was reported to be adenocarcinoma (Fig. 2). It was believed that the lesion was too advanced for surgical procedure; therefore  $^{60}\text{Co}$  irradiation and infusion therapy with Mitomycin C through the both internal iliac arteries was performed.

But he died nine months later. Autopsy revealed that this was the mucoepidermoid cell carcinoma and so-called fistulous carcinoma. Liver and lung metastases were also verified.

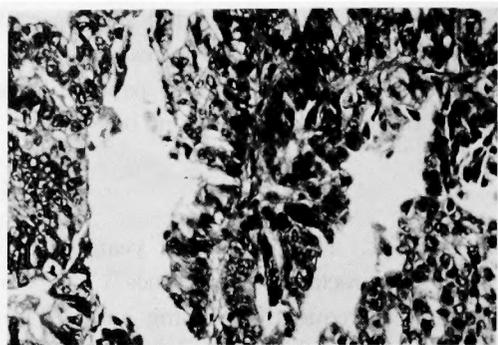


Fig. 2. Histological findings of excised specimen in case 2. Adenocarcinoma (H. E.-staining).

#### COMMENT

The existence of anal fistula for 45 years before an associated carcinoma was detected in one of our cases, this fact emphasizes the possibility of malignancy in any anal fistula. The occasional development of a carcinoma in a fistulous tract is not unknown, yet the question of whether chronic infection and inflammation have any causal relationship with the tumor has not been clearly answered.

One should always keep this possibility in mind in treating a longstanding fistula or sinus and perform biopsy for histological examination in order to exclude the malignant change.

#### SUMMARY

Two cases of long-term fistula-in-ano in which carcinoma later developed, were presented and discussed.

The occurrence of carcinoma in association with anal fistula is very rare but one should keep this possibility in mind in treating a longstanding anal fistula.

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## 和文抄録

### 痔瘻より発生した肛門癌

京都大学医学部外科学教室第2講座 (指導: 木村忠司)

余 昌英・山根 守・石上浩一

直腸癌の中で肛門癌は比較的、稀なもので直腸以下の癌の3~4%を占めるにすぎない。当教室では最近11年余の間に、直腸以下の癌188例中肛門癌は10例にすぎなかつた。

一方、痔瘻は、最も普遍的な疾患の一つであり、ややもすれば、放置されがちである。時に、長期間放置されていた痔瘻の部に癌の発生を見ることが知られている。

1964年 Kline らは、痔瘻に関連せる肛門癌は78例にしかすぎないと報告している。1952年 McIntyre は痔瘻に次の如き条件を見る場合、悪性化の可能性を考

るべきだとした。

即ち、(1) 数年以上も続く瘻孔、(2) 疼痛と硬結の増大、(3) 流出物にムチンを含有するようになる、の3条件である。

我々は最近、45年間存在した痔瘻の部に癌を認められた症例1と25年間存在した、痔瘻の部に認められた肛門周囲癌の症例2を経験したので、これに若干の文献的考察を加え報告した。

本文の要旨は昭和42年10月15日、第22回日本大腸肛門病学会総会にて報告した。