

# Mucoepidermoid carcinoma of the anal canal: A case report and review of the literature

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**Abstract.** Mucoepidermoid carcinoma of the anal canal is a rare tumor. We herein report the case of a 74-year-old male patient with a high-grade mucoepidermoid carcinoma of the anal canal who was treated by local surgical resection and subsequent irradiation. However, the patient succumbed to liver and lung metastases 2 years after the procedure. The characteristic findings of mucoepidermoid carcinoma of the anus remain unclear to date due to rarity of this tumor. Since 1954, when this type of tumor was first described, only 58 cases of patients diagnosed with mucoepidermoid carcinoma of the anus have been reported to date. In this context, a review of the existing English literature on this rare tumor was also performed.

## Introduction

Primary malignant tumors of the anus are histologically diverse and account for only ~2% of all malignancies of the large bowel (1-3). Mucoepidermoid carcinoma of the anal canal, a type of primary malignant tumor, is a rare lesion that is characterized by goblet cell-type mucous cells arranged in solid sheets. To date, only 58 cases have been reported in the literature (4-17). The origin of this tumor is a subject of debate, and its precise histogenesis also remains unclear. We herein present the case of a 74-year-old man with a mucoepidermoid carcinoma of the anal canal.

## Case report

A 74-year-old Japanese male patient was admitted to the Fukuoka Sanno Hospital (Fukuoka, Japan) in January 2013

with complaints of anal pain. Anorexia and vomiting were not reported. Measurement of serum carcinoembryonic antigen and carbohydrate antigen 19-9 revealed that they were below the cut-off levels. Endoscopy revealed a Bormann type II tumor lesion (25x20 mm) in the anal canal (Fig. 1), and biopsy revealed an adenocarcinoma with a squamous cell carcinoma component. Computed tomography revealed enlarged lymph nodes in the right inguinal region and around the right external iliac artery. Resection of the tumor and excisional biopsy of the right inguinal lymph nodes were performed on January 25, 2013, as the patient requested minimally invasive surgery. The patient was discharged on the second postoperative day. The resected specimen was analyzed under an optical microscope at the Armed Forces Institute of Pathology, and pathological examination revealed a high-grade mucoepidermoid carcinoma (Fig. 2A and B), and immunohistochemical staining by p40 and alcian blue (4  $\mu$ m). The blue color indicates the mucin production stained by alcian blue (room temperature, 70 min), and the brown color represents the squamous cells stained by p40 (room temperature -98°C, 180 min) (Fig. 2C and D), with a TNM classification of T2, N3, M0 (stage IIIB). Tumor specimens (2  $\mu$ m) were examined via hematoxylin and eosin (H&E; staining at room temperature for a total of 50 min and analyzed under an optical microscope. The patient underwent radiotherapy in the pelvic and inguinal regions postoperatively; however, he succumbed to lung and liver metastases 2 years postoperatively.

## Discussion

Carcinoma of the anal canal is a comparatively rare disease, constituting only 2% of all malignancies of the large bowel, including the anus (1-3). Some investigators have reported that 4-6.9% of anal carcinomas are diagnosed as mucoepidermoid carcinomas (10,15). However, mucoepidermoid carcinoma of the anus is considered to be a rare lesion. This type of tumor was first described by Kay in 1954, and a total of 58 cases have been reported to date (4-17).

The histogenesis of this tumor has been discussed in the literature. Kay reported that mucoepidermoid carcinoma of the anus arises from 6-8 anal glands, the ducts of which drain into the anal crypts. Fenger and Filip compared the staining

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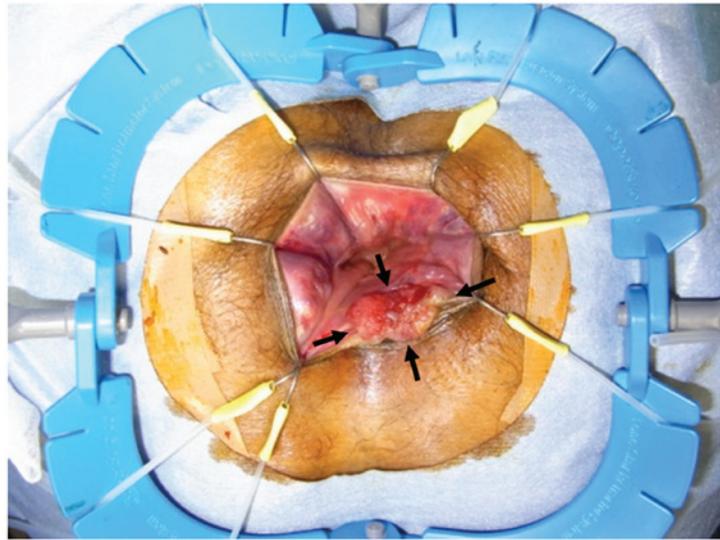


Figure 1. Intraoperatively, a Bormann type II tumor lesion (25x20 mm, indicated by arrows) was identified in the anus.

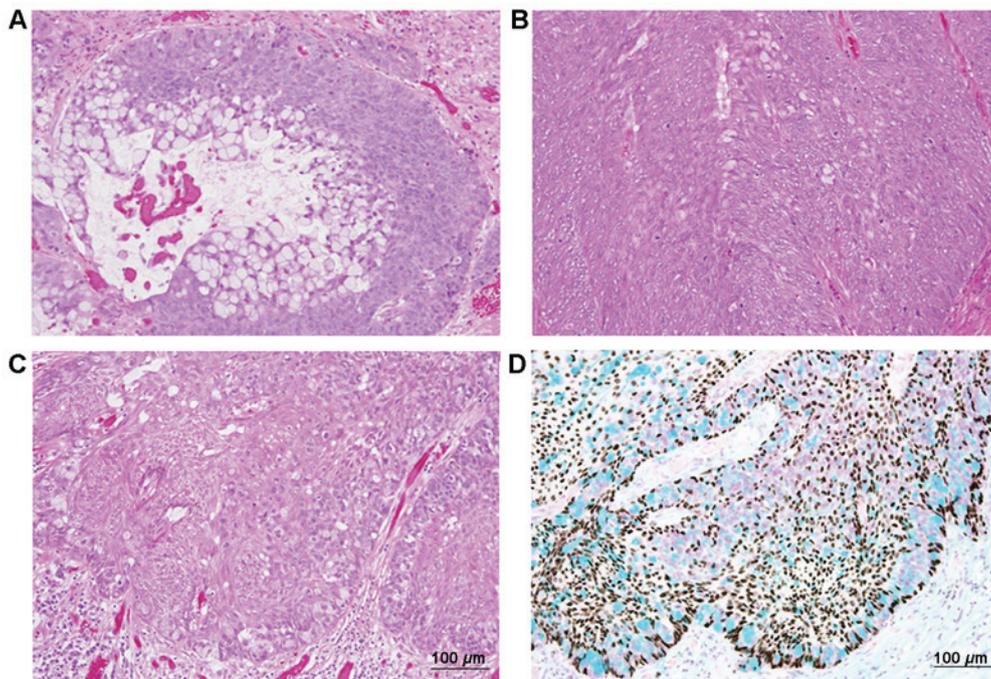


Figure 2. Hematoxylin and eosin (H&E) staining of the tumor specimen revealed the presence of four cell types, namely mucin-producing, squamous, intermediate and clear cells. In the present case, the squamous component was predominant (2  $\mu$ m, room temperature, 50 min). There was no lymphatic or vascular invasion identified in the tumor. (A) Mucoepidermoid (magnification, x100) and (B) squamous cell carcinoma components (magnification, x100). (C) H&E staining in same specimen for D lesion (magnification, x100). (D) Immunohistochemical staining by p40 and alcian blue (4  $\mu$ m, room temperature -98°C, 180 min and room temperature, 70 min). The blue color indicates the mucin production stained by alcian blue, and the brown color represents the squamoid cells stained by p40 (magnification, x100).

characteristics of the mucous component of a mucoepidermoid carcinoma with those of the rectal mucosa and anal glands, and concluded that the staining patterns of mucoepidermoid carcinoma were similar to those of the anal glands (9). By contrast, Morson and Volkstadt and Fenger and Nielsen demonstrated by morphological examination that mucoepidermoid carcinomas arise from the transitional zone, similar to the basaloid and certain other types of squamous cell carcinoma (7,18). The case reported herein falls into the latter category.

Data from 35 cases of mucoepidermoid carcinoma were summarized, including details such as age, sex, treatment

and prognosis (Table I). The mean age  $\pm$  standard deviation was 55.7 $\pm$ 11.3 years, and the male:female ratio was 0.94. All 35 patients had undergone surgery, radiation, and/or chemotherapy. However, the majority of the patients had undergone abdominoperineal resection. By contrast, Papillon and Chassard reported that mucoepidermoid T4 tumors, similar to the epidermoid carcinomas of the anal margin, were suitable for preoperative irradiation and delayed surgery (14). Moreover, the treatment regimens were adjusted according to the depth of tumor invasion or tumor size. The 5-year survival rate in the 35 cases summarized

Table I. Characteristic findings in 35 patients with mucoepidermoid carcinoma.

Characteristics	Values
Age (years)	55.7±7.3
Sex	
Male:female	17:18
Tumor size (cm) <sup>a</sup>	4.09±2.44
Treatment	
APR	22 (62.9%)
Local resection	2
APR + RT	4
APR + chemotherapy	1
Local resection + RT	2
RT	4
Prognosis (survival for >5 years)	11 (31.4%)

Values are presented as mean ± standard deviation, no. (%), or as absolute numbers. <sup>a</sup>Data reported from 12 cases. APR, abdominoperineal resection; RT, radiation therapy.

in the present study was 31.4%, and all the patients had a poor prognosis. Although there have been no reports on the prognosis of mucoepidermoid carcinoma, studies on the prognosis of epidermoid carcinoma reported 5-year survival rates as low as 26 and 32% (8,14).

In conclusion, we herein presented the case of a patient with mucoepidermoid carcinoma of the anus and discussed treatment and prognosis based on previous reports of 35 similar cases. An abdominoperitoneal resection is usually performed in established cases of mucoepidermoid carcinoma of the anus. The prognosis is generally considered to be poorer compared with that of the more common rectal carcinoma.

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### Availability of data and materials

Not applicable.

### Authors' contributions

MY contributed to the design of the present study and the writing of the manuscript. KH, YY, HM, TG and YT made substantial contributions to the acquisition of data. MT contributed to the diagnosis of the mucoepidermoid carcinoma. All authors read and approved the final manuscript.

### Ethics approval and consent to participate

The present study was approved by the Institutional Review Board of Fukuoka Sanno Hospital. Written informed consent was obtained from the patient's family.

### Patient consent for publication

Consent for the publication of data and any associated images was obtained from the patient's family.

### Competing interests

The authors declare that they have no competing interests.

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