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Case Report

Endoscopic Ultrasound-guided Confirmation of Malignancy Arising from Fistula-in-ano

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ABSTRACT

Adenocarcinoma arising from a chronic anorectal fistula (ARF) is rare, and there are no more than 200 cases in literature. We report a case of endoscopic ultrasound-guided biopsy-proven adenocarcinoma arising from long-standing ARF.

KEYWORDS: *Adenocarcinoma, anorectal fistula, endoscopic ultrasound*

INTRODUCTION

An anorectal fistula (ARF) is an epithelialized track that connects anorectum to skin secondary to drainage or rupture of anal gland abscess.[1,2] The true prevalence of anal fistulas is unknown with incidence of an ARF developing from an anal abscess ranges from 26% to 38%.[3] The mean (range) age for presentation of ARF is 40 (20–60) years with twice as common in men compared to women.[1] The common etiologies of ARF are anorectal abscess, Crohn's disease, lymphogranuloma venereum, radiation proctitis, rectal foreign bodies, and actinomycosis. ARF is classified in relationship to the anal sphincter and includes intersphincteric, transsphincteric, suprasphincteric, and extrasphincteric fistulas.[1] Surgical management is the mainstay of therapy. The goal of surgical therapy is to eradicate the fistula while preserving fecal continence.[4]

Adenocarcinoma in long-standing fistula-in-ano is a rare complication with no more than 200 cases reported in literature. It accounts for 2%–3% of all large bowel carcinomas.[5] Due to chronic inflammation of ARF, early diagnosis of rare complication of malignancy can be missed. The origin of fistula associated anal adenocarcinoma is debatable in literature. The clinical hypothesis is that the rectal epithelial cells migrate into the fistula and become malignant,[6] association with Crohn's disease,[7] or heterotopic intestinal cell rests.[8] We report a case of endoscopic ultrasound (EUS)-guided biopsy-proven adenocarcinoma from long-standing fistulain-ano.

CASE REPORT

A 70-year-old gentleman, without any addiction or comorbidities, diagnosed to have fistula-in-ano for the past 40 years, presented with pus and mucus discharge for the past 1 month. He underwent fistulectomy four times before reporting to our hospital. The first surgery was at the age of 30 years with mucopurulent discharge per anus. He had fistulectomy, but the disease recurred back after 5 years, and he underwent redo-fistulectomy another three times for relapse of symptoms with the last surgery being 10 years before the current presentation. He was evaluated in an earlier hospital with normal colonoscopy and benign cells on the resected fistulous tract. He had recurrence of mucopurulent discharge with rectal examination showing surgical scars, induration, and mucopus from two external openings. Colonoscopy was performed for ruling out inflammatory bowel disease, which was negative for the same [Figure 1]. Sonofistulogram and magnetic resonance imaging of the anorectal region revealed complex perianal fistula with intrinsic frond like-polypoid enhancing mass [Figure 2]. EUS was performed using a Pentax linear echoendoscope - EG-3870 UTK connected to a Hitachi Avius estiva ultrasound machine (2012). EUS showed a heterogeneous mass of 3.4 cm × 2.9 cm size with cystic spaces in anorectum. EUS-guided fine-needle aspiration cytology and fine-needle aspiration biopsy [Figure 3] were performed using a Wilson-Cook 22-gauge fine-needle aspiration needle. Cytology and microhistology revealed moderately differentiated mucinous adenocarcinoma [Figure 4]. In view of large tumor and age of the patient, he was

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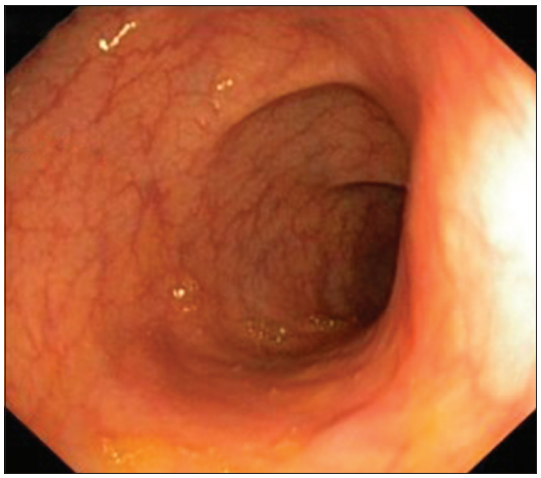

Figure 1: Colonoscopy image showing normal rectal mucosa without any intraluminal tumor

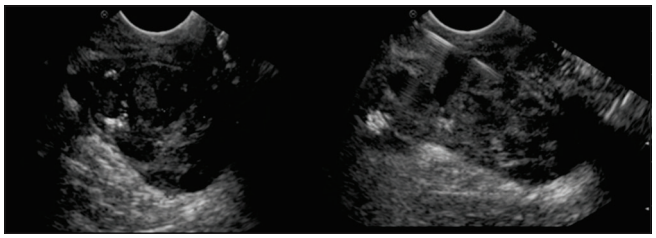

Figure 3: Endoscopic ultrasound image showing 5.1 cm × 2.2 cm heteroechoic mass with solid and cystic areas till 7 cm starting from anal verge and transanal fine-needle aspiration biopsy using a 22-guage Wilson-Cook needle

initiated on chemoradiation to reduce the size of the tumor.

A diagnosis of anorectal carcinoma arising from a former fistula requires not only pathological findings but also knowledge of the clinical course of the patient. For a malignant change to occur in ARF, it should be long standing, without tumor inside anal canal or rectum, and internal opening of fistula in anal canal or rectum should not contain malignant cells.[9] Five clinical features have been reported necessary for a diagnosis of anorectal cancer arising from a fistula: (1) an anal fistula with continuous inflammation for more than 10 years, (2) pain or induration of the anal fistula, (3) mucinous secretions, (4) no evidence of any other primary cancer, and (5) the primary opening being an anal crypt or the anal canal, but etiological relationship lacks surety because of the rarity of the disease.[5]

Mucinous adenocarcinoma of the perianal region oscillates between the pathogenesis of anal and rectal carcinomas causing a diagnostic and therapeutic uncertainty. A high degree of clinical suspicion and histopathological confirmation is required to identify and diagnose.[10] EUS can aid in evaluation of complex fistula-in-ano and ProCore biopsy for excluding

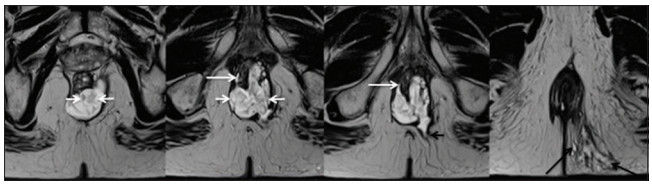

Figure 2: T2-weighted magnetic resonance imaging images showing complex perianal fistula with intrinsic frond-like polypoid enhancing mass. Black long arrows indicate multiple external openings, black short arrow showing one of the tracts, short white arrows showing mass, and long white arrow showing an internal opening in between internal and external sphincter

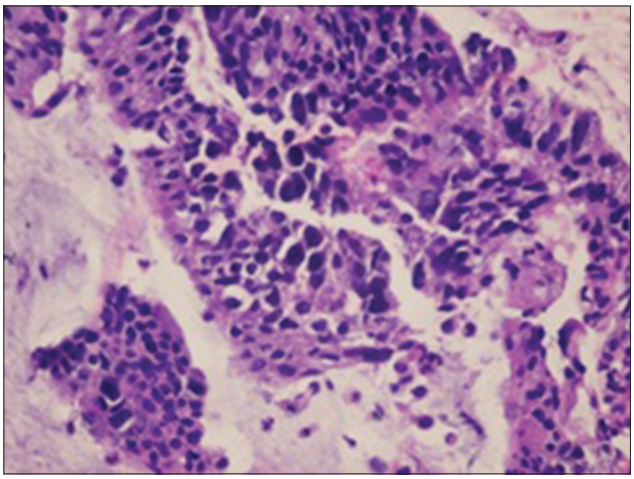

Figure 4: Histopathology photograph showing dilated tortuous glands with lakes of mucin and intervening stroma showing desmoplastic reaction with lymphoplasmacytic infiltration consistent with mucinous adenocarcinoma

malignancy in suspected cases. To our knowledge, this is the first case in literature of EUS confirmed adenocarcinoma arising from fistula-in-ano.

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Conflicts of interest

There are no conflicts of interest.

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