

Anal Amputation and Perineal Flap Reconstruction of an Anorectal Carcinoma Associated With Longstanding Anal Fistulae: Report of A Case

Uzun Sureli Anal Fistül Zemininde Gelişen Anorektal Karsinomun Anal Amputasyon ve Perineal Flep ile Rekonstruksiyonu: Olgu Sunumu

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ÖZET

Amaç: Kronik anal fistül zemininde gelişmiş anorektal karsinom oldukça nadirdir. Bu tür lezyonlar çoğunlukla daha sık görülen benign perianal fistül veya abse ile karıştırılır. Burada kronik perianal fistül zemininde gelişmiş ve agresiv olarak cerrahi tedavi uygulanan bir anorektal kanser olgusu sunulmuştur. Dolayısıyla cerrah, uzun süreli kronik anal fistül zemininde yıllar içinde anorektal kanser gelişebileceğini akılda tutmalıdır.

Anahtar Kelimeler: Adenokarsinom, Anal fistül, flap rekonstruksiyon

ABSTRACT

Purpose: Anorectal carcinoma arising from a chronic anal fistula is extremely rare. Such lesions are usually misdiagnosed for the more common benign perianal abscess or fistula. Herein, we report on a case treated aggressively for anorectal cancer arising from chronic perianal fistulae. Therefore, surgeon should keep in mind that longstanding anal fistula may cause anorectal cancer in years.

Key words: Adenocarcinoma, Fistulae in ano, Flap reconstruction

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Introduction

Anal carcinoma is a rare malignant entity, representing approximately 2-3% of all tumors of the large bowel cancers.¹ It is even uncommon that a carcinoma is found to be arising from a benign fistula-in-ano.² Rosser first reported seven cases in 1934 and identified criteria to define carcinoma originating from anal fistula.³ However, the rarity of this tumor and the lack of sufficient patients for controlled trials have led to a lack of any consensus regarding appropriately diagnostic and treatment strategies.

Case Report

The patient was a 58-year-old male with a history of anal mass and discharging perianal sinus for six years. He had undergone no previous surgery before he was referred to our hospital. At anorectal examination; two external orifices located at the right anterior lateral buttock nearly 3 cm distant from anus and purulent stream were observed (Figur 1). The internal opening of the fistula was identified at the posterior midline in the prone position. A large fibrostenotic area with chronic inflammatory tissue, located at the right side of the anal canal was found. The inguinal lymph nodes were not palpable on both sides. Pelvic magnetic resonance imaging (MRI) and computed tomography (CT) showed both inflammatory tissue and abscess formation in the anorectal region extending to the ischiorectal plan and right side of the buttocks (Figur 2). Rigid sigmoidoscopy showed irregularity and wall thickening in the anal canal



Figur 1. Preoperative view of the patient with perianal opening of the fistulae tract and irregularity in the gluteal region in proctologic prone position.

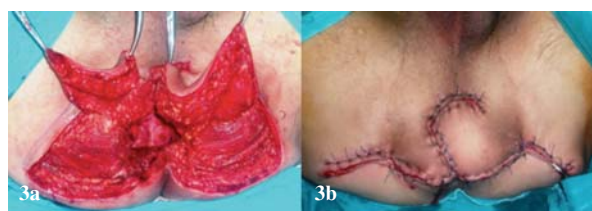


Figur 2. Preoperative MR-imaging of the patient.

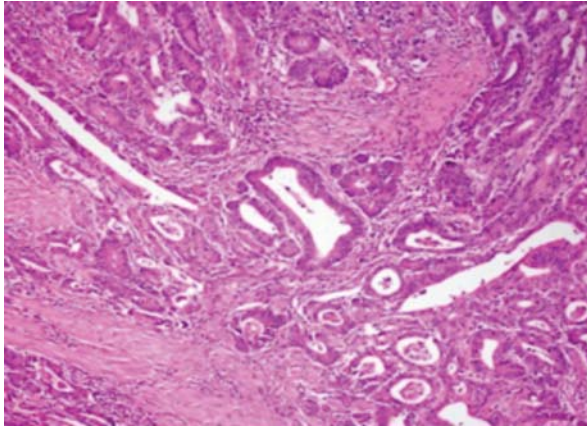
extending to anorectal transitional zone no abnormality. Histological examination of two different biopsy specimens taken from the tract of the fistula with anoderm and anal canal both revealed adenocarcinoma. There was no abnormality on routine laboratory examinations. Tumor markers including carcinoembryonic antigen (CEA), carbohydrate antigen (CA) 19-9 and, AFP, CA 15.3 and CA 125 levels were also within the normal limits. The patient had undergone abdominoperineal resection (APR) + partial coccygectomy and flap reconstruction of perineum (Figur 3 A,B). Postoperative histopathologic examination of the specimen revealed adenocarcinoma (Figur 4). His postoperative period was uneventful and discharged on the 10th day. He had received an adjuvant chemoradiotherapy.

Discussion

Chronic anal fistula is a common disease however perianal adenocarcinoma associated with fistula-in-ano is extremely rare. It has been reported that the frequency in men is twice that in women and nearly 75% of the lesions are found in the lower rectum 4. The pathogenesis



Figur 3 a, b. a. Preparation of anal and gluteal fold flaps. b. Final view of the perineum after reconstruction with bilateral gluteal fold rotational flaps.



Figur 4. Neoplastic glands with cribriform structures infiltrating muscularis propria (H&EX10).

of this disease is still on debate. It is suggested that adenocarcinoma of anal canal may originated from rectum, fistula-in-ano or anal gland. Adenocarcinoma developing in the perianal region is often associated with a longstanding fistula-in ano. The pathophysiology of this condition is in part believed to be secondary to chronic inflammatory changes.²⁻⁴ Rosser established diagnostic criteria for adenocarcinoma originating from the anal-fistula.¹ The fistula should usually antedate the carcinoma by at least 10 years, the only tumor present in the rectum or anal canal should be secondary to direct extension from the carcinoma in the fistula and the internal opening of the fistula should be into the anal canal and not into the tumor itself.²⁻⁵ The most common symptoms are anal pain and perianal mass; anal bleeding and obstruction are generally not reported due to infrequent involvement of the rectal mucosa. Tumor progression can lead to tissue destruction with perianal suppuration and fistula formation. Dukes and Galvin thought these tumors to be a result of malformation or reduplication of the intestinal tract with rest of cells being the source of the tumors.⁶ The origin of carcinoma in areas of chronic inflammation or scarring is thought to be due to the fact that the area in question becomes an “immunological privileged site” in which lymphatic vessels are disrupted and immunologic surveillance against developing metaplastic or frankly neoplastic cells is impaired.⁷ Conventional MRI, CT scanning of whole body with endorectal ultrasonography of anal canal should be done for the diagnosis and for the preoperative staging of the tumor. Colonoscopic

examination should also be helpful for the synchronous tumors in colon.

Currently, surgical resection is considered to be the first choice of curative treatment for anal adenocarcinoma following an adequate excision with clear surgical margin. APR with excision of both ischiorectal fossae and the overlying skin is recommended. Invasion of other organs is often not revealed pathologically after radical resection even when extensive invasion has been detected before or during surgery, because inflammation makes the precise diagnosis of local and lymphatic extension difficult.⁸ The role of radiotherapy in the treatment of perianal carcinoma has not been established; however, careful follow-up and adjuvant chemotherapy or radiotherapy, or both, may prevent local recurrence. The prognosis after APR is generally good, probably because of the propensity of this tumor to be well differentiated and slow growing, and the rarity of metastasis to lymph nodes.⁸⁻¹⁰

Soft-tissue reconstruction following tumor resection is also a challenging problem in perineal region because of the functional, locational, and cosmetic importance of this area. Usually, a large perineal defect is created by radical excision of cancer because of the nature of anal cancer. Several flaps have been designed for perineal reconstruction; each has its advantages together with some disadvantages. An ideal flap for perineal defects should be not bulky, allow stable blood flow, do not cause severe functional or aesthetic alterations in the donor area, and involve only a one-stage operation. The gluteal-fold flap is very useful for various vulvar and buttock reconstructions because it can be adjusted to the required volume. This flap is thin, reliable, easy to elevate, has matched local skin quality, and produces hidden scars on the gluteal fold. All operation scars are restrained to the perineal area and reconstruction can be performed in a single stage. The authors made some modifications on design of this flap and performed bilateral gluteal fold fasciocutaneous rotational flaps for perineal reconstruction following an anal cancer resection. This flap is supplied by underlying fascial plexus derived from perforators of the internal pudental artery and musculocutaneous perforators of underlying muscle.⁹ In this patient, following tumor excision, adequately sized two flaps were designed and lifted with the fascia of the gluteus maximus muscle from the bilateral gluteal

folds. Both flaps survived completely with no complications except for a small perineal wound disruption (Figur 3 A,B). While the donor-site scars, thickness of flap, and degree of flap advancement taken into account, it could be suggested that the gluteal fold fasciocutaneous rotational flaps is a reliable method for reconstruction of perineal defects after tumor resection. In conclusion; fistula-associated anal carcinoma is an uncommon malignant transformation of chronic fistula-in-ano. Conventional radiologic methods as MRI, CT and endoanal ultrasound provide diagnostic information

on patient with this suspicious inflammatory condition. Although radical surgery for this tumor with abdominoperineal resection remains the primary surgical treatment of choice, combined chemoradiotherapy may be appropriate for these patients with promising results. In our practice, after aggressive surgical amputation of the tumor, the gluteal fold fasciocutaneous rotational flaps have proven to be useful for perineal reconstruction, especially at the point of donor-site scar, flap thickness, and degree of flap advancement.

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