

CASE REPORT

Mucinous Cystadenocarcinoma in Chronic Anorectal Fistula: Clinical and MRI Characteristics

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Abstract

A 54 years old Libyan man presented with a long history of perineal mass, induration and passage of pus and purulent secretion; sometimes mixed with blood. It became increasingly painful over a short period of a few days prior to presentation. He had had a fistula-in-ano for ten years treated surgically 8 years previously. His past medical history included type 1 diabetes mellitus. He underwent digital rectal examination under anesthesia which disclosed edematous external skin tags, with induration of the posterior margin of the anal canal, multiple biopsies and scrapings of the fistulous track were taken. Histological examination revealed mucinous adenocarcinoma. Colonoscopy was done and showed no abnormality. Clinical and MRI characteristics are illustrated and discussed.

Key words: Mucinous adenocarcinoma; Anorectal fistula, MRI

Introduction

Mucinous adenocarcinoma in association with chronic fistula-in-ano is a rare entity. Diagnosis is often difficult (1,2). This leads to severe problems concerning treatment and prognosis. Biopsy of the external openings of the fistulous tracts is not conclusive and very often misleading as the tissue taken is very superficial and only reveals an inflammatory reaction, especially when scarring and fibrosis exist (2,3). Magnetic Resonance Imaging (MRI) has been considered the most accurate preoperative technique for classification of fistula-in-ano. It is particularly useful for evaluation of the primary track and extensions and for distinction from pilonidal sinus disease (2,4,5). A few cases of mucinous adenocarcinomas arising from fistula-in-ano detected by MRI have been described in the literature. We describe an illustrative case of mucinous adenocarcinomas complicating a fistula-in-ano and demonstrate its MRI characteristics.

Case Report

Clinical history

A 54 years old Libyan man presented with a long history of perineal mass, induration and passage of pus and purulent secretion; sometimes mixed with blood (Figure 1). It became increasingly painful over a short period of a few days prior to presentation. He had had a fistula-in-ano for ten years treated surgically 8 years previously. His past medical history included type 1 diabetes mellitus. He underwent digital rectal examination under anesthesia which disclosed edematous external skin tags, with induration of the posterior margin of the anal canal, multiple biopsies and scrapings of the fistulous track were taken. Histological examination revealed mucinous adenocarcinoma. Colonoscopy was done and showed no abnormality.

The MRI protocol

MRI using a phased-array pelvic coil was performed using Philips Intera 1.5, MRI Machine (Philips Healthcare, DA Best, The Netherlands). The imaging process was supervised by a specialist radiologist. The long axis of the anal canal was identified by using a midline sagittal localized image. Using this image for guidance, axial, sagittal, and coronal T2-weighted fast spin-echo, axial T1-weighted spin-echo images were performed, and coronal and sagittal T1-weighted spin-echo with fat suppression images were obtained immediately after intravenous bolus injection of 0.1 mmol/kg body weight of Gadopentetate dimeglumine (Magnevist, Schering, Berlin, Germany).



Figure 1. Large ulcerating left perianal mass lesion

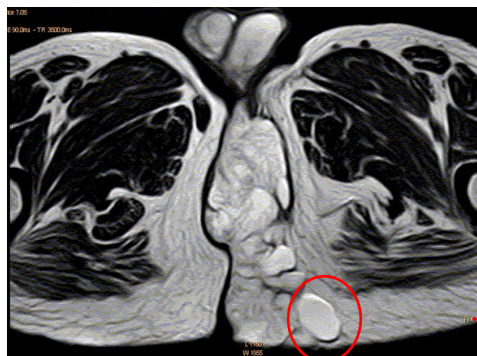


Figure 3. Axial T2 sequence demonstrate an area of homogenous sharp hyperintense signal in the left side of the perineal sub-cutaneous fat tissue (circle), this area lacks a fibrous capsule which is indicating mucinous pool.

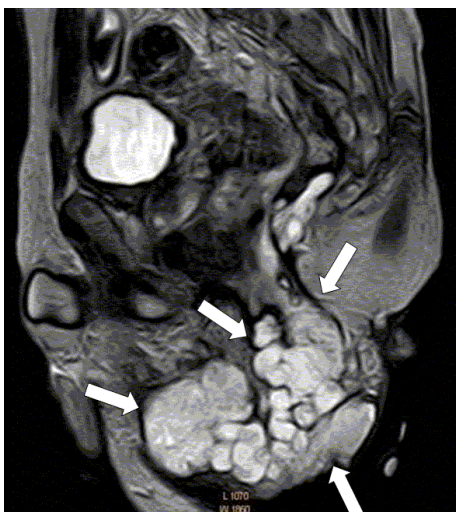


Figure 2. T2 sequence in sagittal planes, shows well delineated perineal mass lesion (arrows) outlined by fibrous capsule exhibit lobular outer contour with hyperintense signal character.

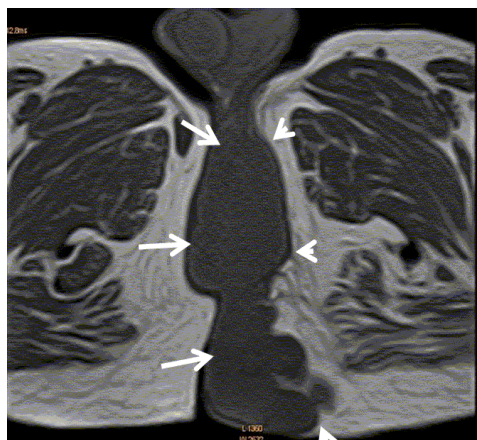


Figure 4. Axial T1 sequence reveals well defined left paramedian perineal sub-cutaneous mass lesion (arrows) exhibiting homogenous hypointense signal surrounded by thin capsular deep hypointensity (arrow head).

The MRI Characteristics

The resulted image shows the following radiological characteristics:

a) Lobulated left sided perineal sub-cutaneous mass lesion measuring about 10 cm in its longest diameter, surrounded by thin fibrous capsule with T2 hyperintense signal character (Figure 2).

b) Loculated area of sharp T2 hyperintensity not surrounded by fibrous capsule which indicates presence of mucin pool (Figure 3).

c) In T1 sequence the lesion appears homogenously hypointense with deeper hypointensity of the fibrous capsule (Figure 4).

d) T2 Fat Sat clearly demonstrates multiple thin internal septations that appear hypointense traversing the hyperintense tumoral bulk with the surrounding fibrous capsule also appear hypointense while the mucin pool exhibit homogenous sharp hyperintensity (Figure 5).

e) Well defined fistula tract connecting the lesion with the tumor mass, appears as a tubular structure surrounded by fibrous capsule, containing fluid signal (hypointense in T1 and hyperintense in both T2 and T2 Fat Sat sequences), surrounding the entire circumference of the anal canal (Figure 6 a,b,c,d)

f) After intravenous Gd-DTPA administration the lesion perse exhibits mild heterogeneous contrast enhancement with intense peripheral and internal septal enhancement, while the wall of the fistula shows sharp enhancement

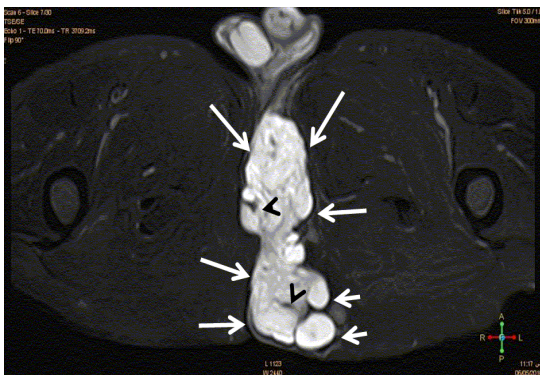


Figure 5. Axial T2 Fat Sat sequence showing the left side perineal sub-cutaneous mass lesion with different degrees of hyperintensity (long white arrows) tumor mass, (short white arrow): mucin pool, containing multiple internal hypointense septae (black arrow head).

pattern (Figure 6a, 6b).

g) Associated findings raising the possibility of the malignancy include enlarged pelvic lymph nodes; the largest measuring about 3 cm in diameter (Figure 7).

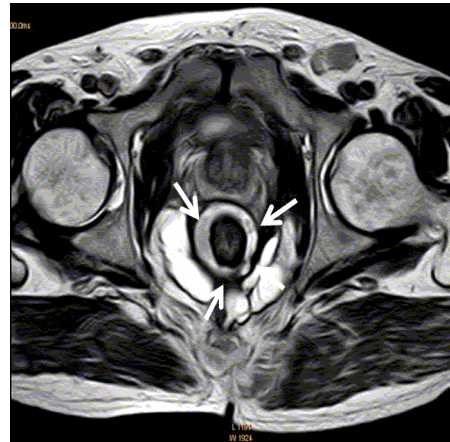


Figure 6a. Axial T2 sequence revealing intra-pelvic extension of the fistula with complete encasement of the anal circumference

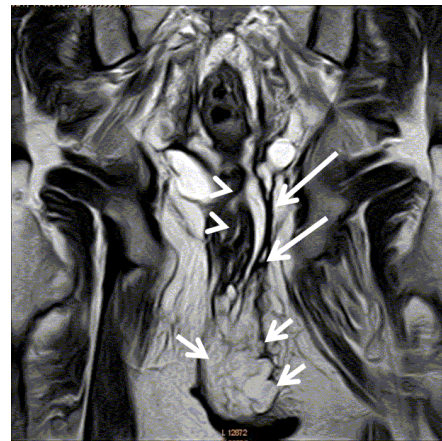


Figure 6b. Coronal T2 sequence clearly demonstrating the left side peri-anal fistula (long white arrows) that connecting the large perineal mass lesion (short white arrows) with the anal canal (arrow heads).

Discussion

The anal intramuscular glands are simple tubular, sometimes branched, glands that open into the anal canal via the crypts of Morgagni. Histologically, they are lined by simple or cuboidal epithelium and may penetrate the internal and external sphincter muscles as well as the subcutaneous fat. These glands being modified sebaceous glands, the secretions are formed from sloughed epithelium. The

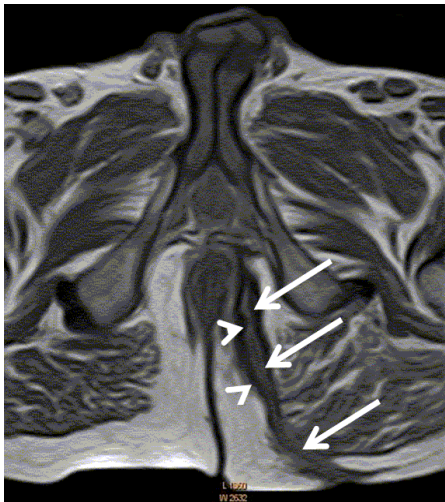


Figure 6c. Axial T1 sequence showing hypointense fistula tract (white arrows) surrounded by thin fibrous capsule (white arrow head) running in the left side perineal sub-cutaneous tissue.

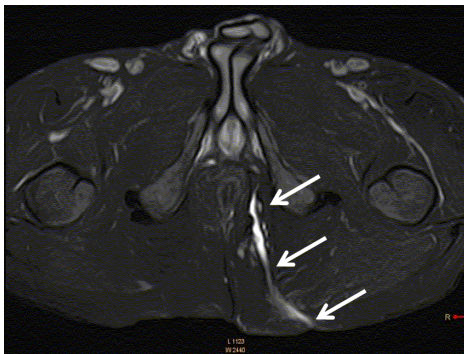


Figure 6d. Axial T2 Fat Sat sequence showing sharp hyperintense fistula tract (arrows) in the left side perineal sub-cutaneous tissue

function of the anal glands is to produce mucus, which is

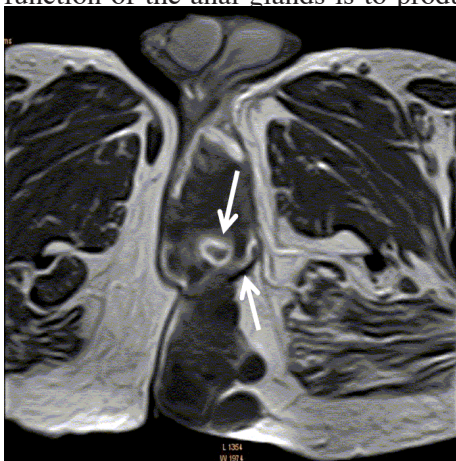


Figure 7a. Axial T1 sequence after intravenous Gd-DTPA administration revealing intense internal septal enhancement (white arrows), with mild heterogeneous enhancement of the rest tumor bulk

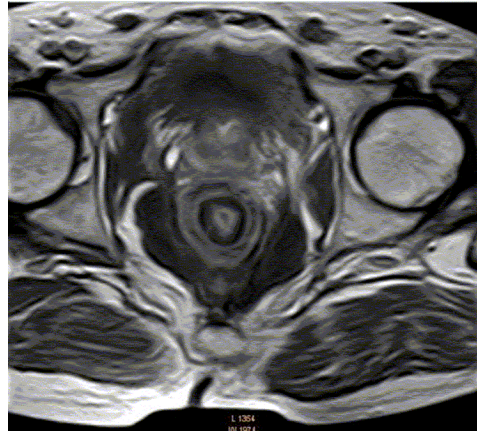


Figure 7b. Axial T1 sequence after intravenous Gd-DTPA scan administration showing intense fistulous fibrous capsular enhancement



Figure 8. Axial T2 sequence depicting enlarged pelvic lymph nodes.

conveyed via the anal ducts to the crypts for lubrication. Because these glands extend downward into the tissues and outward into the sphincter musculature, the importance of these ducts and glands with regard to the spread of infection is considerable (14). Jones and Morson described the origin of mucinous adenocarcinoma in anorectal fistula (14,15). They suggested that the fistulous tracks are congenital duplications of the lower end of the hind gut lined by rectal mucosa that is prone to malignant change to mucinous adenocarcinoma (14,15). Other basis of the mucinous adenocarcinoma in an anorectal fistula may be chronic pancolitis, Crohn's disease with high inflammatory activity, or persistence of chronic fistulas and stenosis that seem to trigger malignant transformation (14,16). Anal fistula may also arise from infected anal glands, and it may be speculated that chronic inflammation could also induce anal gland epithelium to undergo neoplastic change

Table 1. MRI criteria of Mucinous Cystadenocarcinoma

Radiological Criteria	Description
Markedly hyperintense fluid on T2-weighted images	Signal intensity similar to or brighter than perirectal fat (2,8).
Enhancing solid components	Areas with a major axis at least 5 mm long located in the fluid or mucin pool and with contrast enhancement after IV administration of Gadolinium chelate
Meshlike internal enhancement	Formed by cells, cords, and vessels (2,10,11)
Fistula between the mass and the anus	A high signal-intensity communication between the mass and the anus assessed on T2-weighted images, which is a characteristic finding of mucinous adenocarcinoma arising from fistula-in-ano (2,7)
Included fluid collections without a thick fibrous capsule	Reflects the mucin pool of mucinous adenocarcinomas (2,8)
Contrast enhancement of peripheral structures or peritumoral areas	Assessed on the fat-suppressed contrast-enhanced T1-weighted images which may be a reliable finding reflecting the invasion of cancer cells.
Regional areas of lymph node enlargement	Perirectal and internal iliac lymph nodes greater than 10 mm in the short axis and inguinal lymph nodes greater than 15 mm in the short axis (2,6,7,14).

(7,14,17).

MRI accurately shows the perianal anatomy and is highly valuable in the preoperative assessment of fistula-in-ano (2,10). It can be diagnosed by its characteristic MRI findings (Table 1). Our case demonstrated all these criteria together with signs of invasiveness. With special consideration to the active inflammation within the fistula-in-ano or abscess which is similarly enhances the contrast material in peripheral structures or peritumoral areas, regardless of the presence or absence of mucinous adenocarcinomas. Therefore, MRI might be particularly useful after inflammation subsides. Regional lymphadenopathy which is a nonspecific finding may be a helpful sign of locally advanced disease. However, correlating the associated MRI findings and clinical features should help narrow the diagnostic possibilities.

In conclusion, cases of mucinous adenocarcinoma in anorectal fistula are rare. If surgical treatment for perineal abscess or anorectal fistula is not successful for a long time, mucinous adenocarcinoma should be suspected. Prognosis after surgical mucinous adenocarcinoma treatment is good if the condition is diagnosed and treated early (6,7,15,18,19).

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