



Fistula-in-ano Extending to the Thigh

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ABSTRACT

Fistula-in-ano tracts are usually short and open around the perianal region but it is uncommon to have tracts opening beyond this region. Hence, complex fistulas remain a therapeutic challenge and are often linked to recurrence. We describe herein an unusual fistula-in-ano extending to the mid-thigh posteriorly which was managed successfully by fistulectomy and secondary healing.

Keywords: Complex fistula, fistula-in-ano, fistulectomy, thigh

Introduction

A fistula is an abnormal communication between two epithelialized surfaces.¹

A typical fistula-in-ano usually consists of a tract with an internal opening in the anus or rectum and external opening(s) on the perianal skin.² It is quite uncommon for the fistula to extend beyond the perineal region.³ The majority of anorectal fistulas are cryptoglandular in origin. Other causes include trauma, Crohn's disease, malignancy, radiation, or unusual infections (tuberculosis, actinomycosis, and chlamydia).²

Diagnosing a fistula-in-ano can be complicated in the presence of unusual clinical symptoms.⁴ This happens because the fistula can traverse unusual courses (complicated tracts, curved tracts, multiple openings or those which take a circuitous path to the anal canal) causing diagnostic dilemma.^{2,4} The need for treatment is because of persistence as spontaneous healing rarely occurs. This may be due to epithelialization of the fistula tract, which prevents the fistula from closing.²

Due to different etiologies and risk factors, the approach to management varies.¹ It is necessary to have a patient-oriented approach depending on the complexity. This report

highlights a rare presentation of a common condition - fistula-in-ano and its management.

Case Report

A 42-year-old man from Dar-es-Salaam presented to us with three weeks history of pus discharge from the right thigh, which was gradual in onset. The pus was foul smelling, blood stained and approximately 10 cc per day. It was associated with swelling of the thigh and sharp pain around the anal orifice, which was severe on defecation. The pain was relieved by use of over-the-counter analgesics. The condition was also associated with intermittent low-grade fever. The patient reported the condition to have persisted for more than eight years during which he has undergone multiple surgeries on the same limb at various centers but without success.

In 2012, the patient underwent incision and drainage at a regional level hospital during which 1500 milliliters of pus were drained. Thereafter, the patient attended and was scheduled for surgery on multiple occasions at a tertiary level hospital; however, the surgery never took place.

There was no history of abdominal pain, distention, constipation or diarrhea. There was no history of inflammatory bowel diseases or diabetes. He was hypertensive on regular



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oral therapy. The patient did not consume alcohol or use tobacco. He was not addicted to any prescription or recreational drug. There was no significant personal or family history of chronic diseases. The patient denied being on any medication.

On general examination, the patient was overweight. Examination of abdominal systems was unremarkable, apart from digital rectal exam which was tender and hence not completed. Local examination revealed a scar from previous incision and drainage in the posterior proximal half of the right thigh, which was oozing seropurulent effluent. There was another scar in the distal third of the thigh posteromedially. No overt sinus or fistula was noted around the perianal region (Figure 1).

The patient underwent colonoscopy, which was unremarkable, and magnetic resonance fistulogram which revealed a long track arising from the posterior external anal sphincter at six o'clock tracking into the right gluteus muscle and inferiorly to the posterior fascia between the semitendinosus and biceps femoris muscles. The track exited through the subcutaneous tissue of the right proximal posterior thigh. The fistula track measured approximately twenty-two centimeters in length. Features suggested fistula-in-ano with the external opening at the right proximal posterior thigh (Figure 2).

Hematological and biochemical parameters of the patient were within normal range. Serology for hepatitis A, B and C, and human immunodeficiency virus were negative.

The patient was scheduled for surgery and the following were encountered intra-operatively: methylene blue dye injected at the discharging site in the posterior mid-thigh was noted to come from the anal os about 20 mm from the anal verge. A 5 Fr ureteric catheter was inserted but was unsuccessful beyond three centimeters. A hemostat

forceps was inserted and a longitudinal incision made from the caudal to the cranial end where the fistula formed a T-shaped tract; one leading towards the anal canal at two centimeters from the anal verge and another laterally about three centimeters (Figure 3).

The fistula was determined to be a Park's type 1 intersphincteric fistula. The fistula tract was opened and subsequently excised, measuring thirty-two centimeters. The wound was laid open, washed thoroughly, and packed with paraffin dressing (Figure 4).

The excised tract was sent for histology and revealed fragments of fibrous wall infiltrated by mononuclear inflammatory cells and foreign body giant cells, suggestive of chronic granulomatous inflammation (Figure 5).

Post-operatively, the patient improved significantly with the wound healing rapidly by secondary intention (Figure 6). He underwent daily dressings, followed by sitz-bath and was kept on oral flucloxacillin 500 milligram per oral three times a day for fourteen days, and thereafter dressing only was performed.

Discussion

Fistula-in-ano is one of the oldest pathologies in mankind and was first analyzed by Hippocrates.⁵ The incidence rates for men and women are 12.3 and 5.6 in 100,000 population respectively, though it's difficult to make accurate estimations due to the embarrassing symptoms.^{6,7} Fistula-in-ano is a chronic manifestation of an acute perirectal abscess that when ruptured or drained, forms an epithelialized tract connecting the abscess in the anus or rectum to the perineal skin. It is estimated that 40% of peri-anal abscesses will be accompanied or preceded by fistula.^{2,7} These patients often present with recurrent malodorous perianal discharge, pruritus, recurrent abscesses and perianal pain.² From the

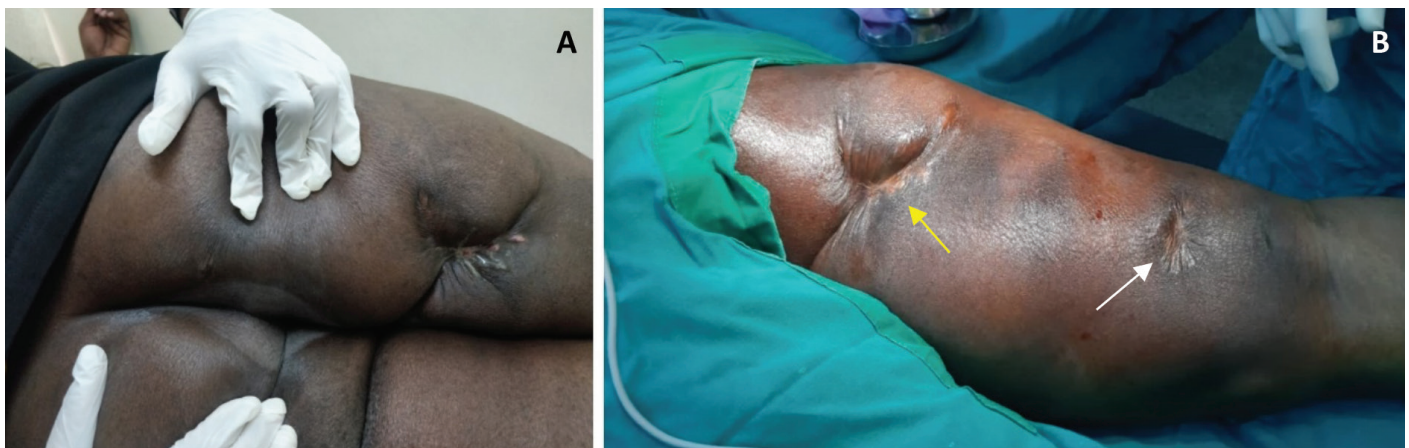


Figure 1. Pre-operative images showing scars from previous incision and drainage (A). Site of active pus discharge where the contrast was injected (yellow arrow), a healed scar with a tract underneath that was not stained by contrast and as such, did not appear on the magnetic resonance imaging (white arrow) (B)

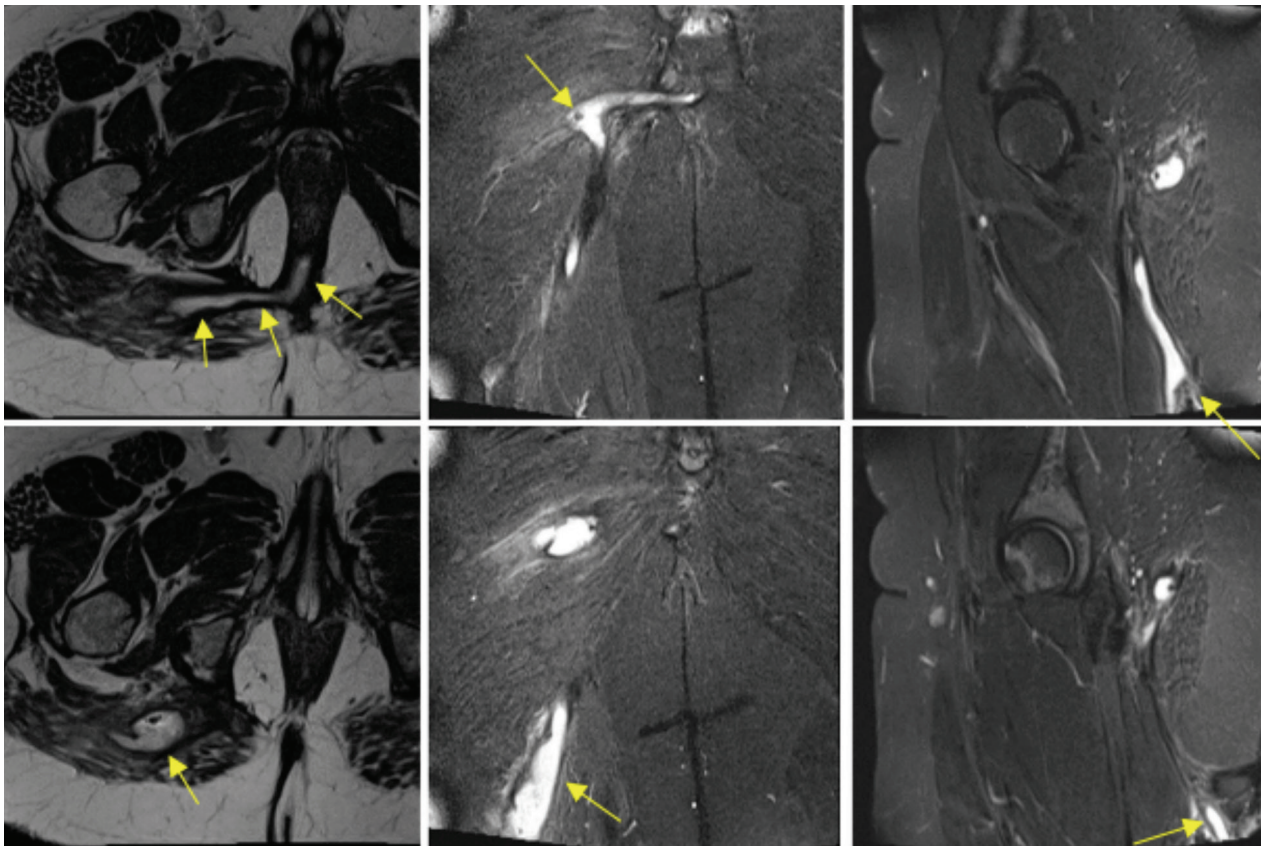


Figure 2. Magnetic resonance fistulogram (T2 axial, T2 fat sat coronal and sagittal images) shows a long sinus track arising from the posterior external anal sphincter at 6 o'clock, tracking into the right gluteus muscle and inferiorly to the posterior thigh muscle fascia (between the semitendinosus and biceps femoris muscles). The sinus track exits through the subcutaneous tissue of the right proximal posterior thigh. The tract measured approximately 22 centimeters in length

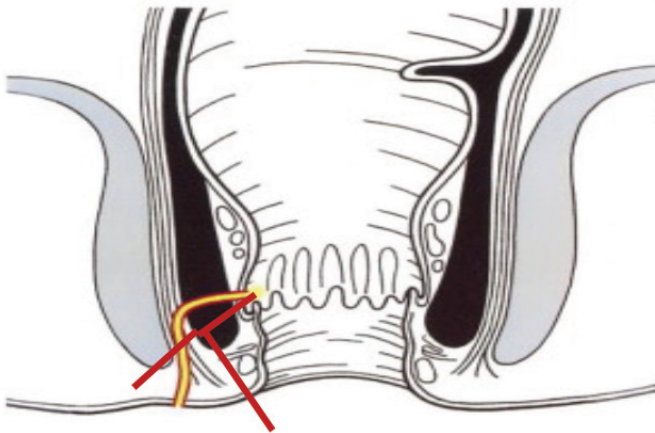


Figure 3. T-shaped tract (red); one leading towards the anal canal and the other ending as a blind sinus laterally (image courtesy: science direct)

index case, it is evident that the patient also presented with continuous recurrent discharge from the external site (thigh), and had a history of low-grade fevers, intermittent perianal pain and underwent several incisions and drainage of the abscess.

A typical fistula tract has an internal (primary) opening in the rectum or anus and an external (secondary) opening on the perirectal skin.² In contrast, our patient presented with an unusually long tract with the secondary opening on the thigh, causing a diagnostic dilemma similar to that reported by Ertekin et al.² in their case report. The most common taxonomy used to classify fistula-in-ano is the Park's classification; inter-sphincteric (70%), trans-sphincteric, supra-sphincteric, and extra-sphincteric (1%). This is described by the course of the fistula tract in relation to the anal sphincters. The commonest type also has the best prognosis as also revealed by rapid recovery of our patient. The majority of fistulas are cryptoglandular in origin, as in the index case. Other causes can be trauma, Crohn's disease, malignancy, radiation, infections such as tuberculosis or chlamydia, but all of these risk factors were ruled out in our case.^{6,8}

Management of fistula-in-ano varies according to its complexity. For complex fistula, a Seton suture is advocated to allow drainage of sepsis and a mature tract of fibrous tissue to develop. After this, definitive fistula treatment is considered from the various options described

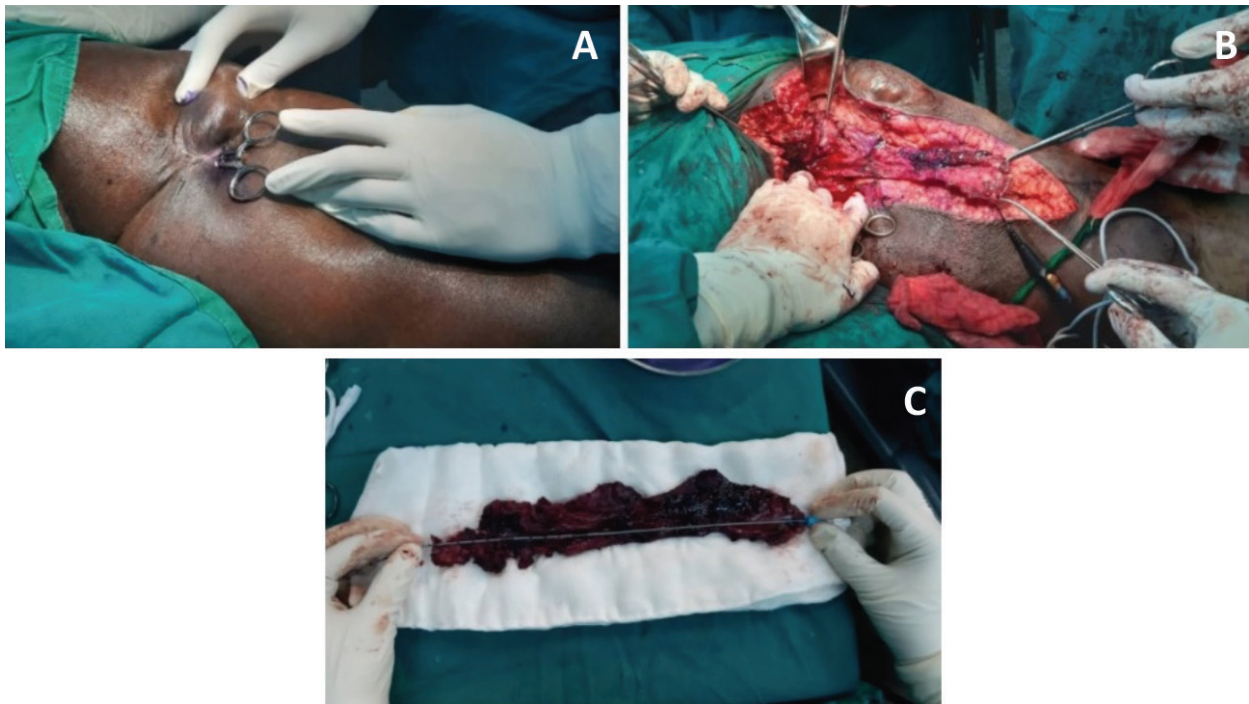


Figure 4. Intraoperative images showing a straight hemostat inserted in the distal os of the fistula (A), the entire length of the tract laid open (B), and the length of the excised tract tissue (C)

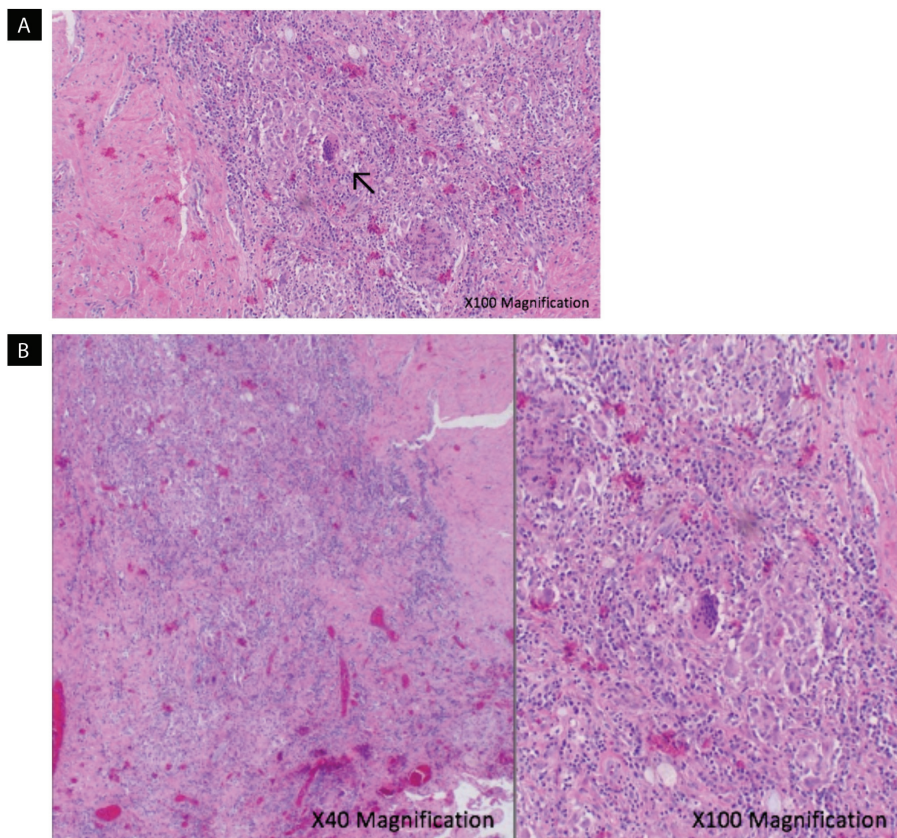


Figure 5. (A) hematoxylin and eosin-stained sections showing focus of dense chronic inflammation consisting of foreign body giant cell indicated by an arrow (↖) raising concern about granulomatous inflammation. (B) A and B sections from the wall of the cystic lesion of the fistula, exhibiting extensive mononuclear inflammatory cells with some giant cells, again raising concern about granulomatous inflammatory process (conventional hematoxylin and eosin staining)

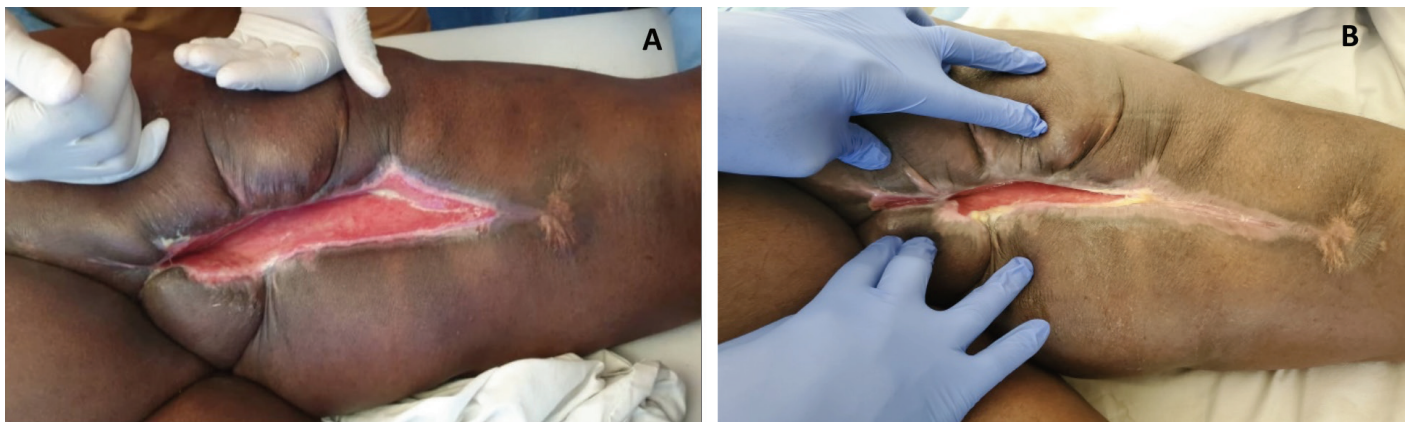


Figure 6. Healing by secondary intention six weeks (A) and eighteen weeks (B) post operation

in the literature.⁷ Keogh and Smart⁷ proposed the use of radiofrequency ablation after the use of Seton suture to minimize tissue destruction, as seen in fistulectomy and fistulotomy, and this would be less painful. Due to high recurrence rates and complications from various surgical techniques, other authors are still attempting to identify optimal methods, such as the use of Ayurvedic *Kshara Sutra* with a reported success rate of 96.6%.⁹ Fistulotomy has been described to be superior to fistulectomy. However, we opted for the latter since the fistula tract had significant fibrosis, which would hinder healing by granulation and could potentially be a source of retained infection due to accumulation of debris. We wanted to leave fresh tissues free from chronic inflammation to promote healing. Moreover, the history of recurrence favored fistulectomy over fistulotomy. The risk of incontinence from fistulectomy was assessed to be negligible as more than two thirds of the fibers were spared. Other surgical modalities include advancement flaps with or without fibrin sealant.² Ertekin et al.² opted for a more conservative approach using 1% silver nitrate solution to irrigate the fistula but with a long healing period.

Conclusion

Fistula-in-ano is not a completely innocuous disease and patients should be counseled to seek prompt and adequate treatment. Although various treatment modalities have been described, no treatment is considered optimal.

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Ethics

Informed Consent: Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: M.T., A.D.M., K.C., Concept: M.T., J.L., K.C., Design: M.T., J.L., Data Collection or Processing: M.T., A.M., A.D.M., Analysis or Interpretation: M.T., P.A., A.S., Literature Search: M.T., J.L., A.M., Writing: M.T., J.L., A.M.

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