



ANAL FISTULA WITH MULTIPLE EXTERNAL OPENINGS: A CASE REPORT SUCCESSFUL TREATMENT THROUGH FISTULECTOMY AND SPHINCTEROPLASTY INTEGRATION

Medical Education

**Dr Parag
Nawalkar**

Consultant, Pristyn Care, Pune, Maharashtra, India

Dr Mohd

**Azharuddin Azim
Attar**

Consultant, Pristyn Care, Pune, Maharashtra, India

**Dr Vaibhav
Kapoor**

Co-founder, Pristyn care, Gurgaon, Haryana, India

**Mitesh Mohan
Hood***

AGM-Medical Excellence, Pristyn care, Gurgaon, Haryana, India *Corresponding Author

ABSTRACT

Background: Complex anal fistulas with multiple openings are rare and difficult to treat. Hence, it is necessary to choose appropriate surgical treatment and ensure the sphincter reconstruction to avoid postoperative complications. **Method:** In this study, a 23-year-old man was presented with fistula-in-ano with three external openings and no internal opening. Confirmation of the findings was obtained through magnetic resonance (MR) fistulography. Treatment consisted of fistulectomy in conjunction with laser ablation. External sphincteroplasty was conducted using vicryl 2-0 absorbable sutures. **Results:** The patient was discharged the next day after surgery and followed up for 8 weeks without experiencing any postoperative complications. **Conclusion:** Combining fistulectomy with sphincteroplasty could offer a more effective and safer treatment approach for complex fistulas.

KEYWORDS

Fistula-in-ano, Magnetic Resonance Fistulography (MR fistulography), Fistulectomy, Sphincteroplasty, Case report

INTRODUCTION

Fistula-in-ano is a prevalent yet debilitating condition, exhibiting a higher incidence in males. Studies report a prevalence of 10-30 cases per 100,000 individuals in men, with a male-to-female sex ratio of 2 to 3.¹ An anal fistula is a tunnel-like connection between the anus, and the nearby skin, formed when an abscess near the anus bursts.

This condition often fails to heal properly, leading to ongoing inflammation and drainage.² Beyond the typical gland infection, anal fistulas can also proliferate from tuberculosis, lymphogranuloma inguinale, and inflammatory bowel diseases like Crohn's and ulcerative colitis.³

Infections spreading subcutaneously (under the skin) may lead to the formation of multiple openings, which can complicate the condition.⁴ Complex anal fistulas with multiple openings are rare^{5,6} and tend to co-exist with some other comorbidity such as tuberculosis.⁵

For fistulas with several openings, surgery is the most effective approach. There are various surgical techniques available, including anal fistula plugs,⁷ ligations of the internal tract, glue injections, flap procedures,⁸ and minimally invasive video-assisted surgery.

This case report presents a unique instance of an anal fistula with three external openings, where the internal opening was not identified despite magnetic resonance imaging (MRI) evaluation. This case report outlines the treatment approaches employed for managing complex anal fistula.

Case Report:

A 23-year-old male patient presented to our outpatient department with chief complaints of skin irritation and pus discharge around the anus for 2 weeks. No complaints of abdominal pain or frequent stools were reported. The patient denied any history of passing mucus or blood via the rectum.

The patient's medical history revealed no prior surgical procedures or significant past illnesses. To alleviate the symptoms, the patient was taking some ayurvedic medication. There was no history of diabetes, tuberculosis, hypertension, or bronchial asthma in the family.

The patient was extremely comfortable during clinical observation. No symptoms of abdominal inflammation, tenderness, redness, or drainage were observed.



Fig1A: Physical examination site of the patient showing the fistula-in-ano with three external openings.



Fig1B: Healed surgical site after 8 weeks follow-up.

Physical examination of the patient revealed a fistula-in-ano with three external openings around the anus at 4 o'clock, 5 o'clock, and 6 o'clock positions. (Fig 1) Upon gentle palpation of the aberrant area, fluctuance was detectable, causing increased pain for the patient. The distal-most opening was around 6 cm from the anus and no internal opening was detected. The patient was advised to have a Magnetic Resonance fistulography to better understand the underlying position. (Fig 2) The Fistulogram revealed a perianal sinus with three external

openings and no obvious internal opening. No internal collection or secondary tract was identified. Based on these findings, a fistulectomy was suggested.

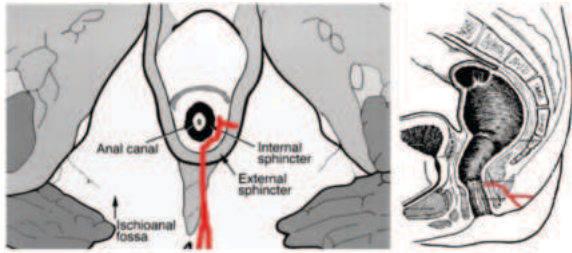


Fig 2: MR fistulography revealing the anatomy of fistula-in-ano, providing comprehensive insight into its structure and path.

After receiving consent, the patient was evaluated in the lithotomy position and the treatment was initiated. Under spinal anesthesia, methylene blue, and hydrogen peroxide were injected through one of the external openings to clean the fistula tract and locate the internal opening. All 3 openings were interconnected in a Y-shaped tract (**Fig 2**) with no obvious internal opening. Tracts were curetted till the intersphincteric space was reached and the dye was again injected. No internal opening was found, and the tract was ablated with a radial fiber coupled with a neon diode laser after cleaning up to the intersphincteric space. To avoid further anal incontinence external sphincteroplasty was done with vicryl 2-0 absorbable sutures. The patient was discharged the next day after surgery. The post-operative period in the hospital was uneventful. The patient was followed up for 8 weeks post-surgery. All the wounds healed within 4-6 weeks. (**Fig 1B**) The patient was healthy and fine.

DISCUSSION:

Fistula-in-ano, characterized by multiple external openings and the absence of an internal opening, represents a rare yet intricate condition.¹⁰ Managing such complex fistulas poses significant challenges due to the risk of recurrence and potential impairment of anal continence.¹¹ The fundamental principles guiding surgical management of anal fistula involve accurate identification of the fistula tract, complete removal of the diseased tissue, and preservation of anal sphincter function.¹⁰ Preoperative diagnosis and structural identification of anal fistulas play a crucial role in initiating appropriate treatment. The popularization of intracavitary ultrasound and MRI has enhanced the accuracy of fistula diagnosis by providing detailed anatomical information.¹¹ MRI, the gold standard for anal fistula diagnosis, offers precise visualization of the internal opening and the course of the fistula tract, facilitating optimal treatment planning and improving patient outcomes.¹¹

In this case, the patient was presented with three external openings on the left side of the perianal region. To further delineate anatomical features and exclude any internal pathology, an MR fistulography (**Fig 2**) was recommended. The examination unveiled an intersphincteric sinus tract measuring 10.8 cm in length, situated on the left side of the perianal region. Its external opening was pointed at the 6 o'clock position in the perineum, displaying a maximum thickness of 7 mm. The tract followed an anterosuperior course in the left paramedian location, extending approximately 8 cm and reaching the 6 o'clock position at the level of the puborectalis sling. Subsequently, it traversed into the intersphincteric region, spanning from the 6 to the 3 o'clock position for about 2.8 cm at the same level. (**Fig 2**) No obvious internal opening could be detected.

Several surgical techniques, such as fistulectomy, seton placement, and advancement flap procedures, have been documented for treating fistula-in-ano.⁸ However, determining the optimal management strategy relies on factors specific to each patient and the anatomy of their fistula tract.¹¹ In instances where multiple external openings exist without a clear internal opening, a combined approach involving complete excision of the tract alongside sphincteroplasty may present as a more effective and safer treatment option. This integrated method contributes to achieving definitive closure while simultaneously lowering the likelihood of recurrence and postoperative incontinence.¹² Since no internal opening could be identified even after the MR imaging, the patient was recommended for fistulectomy. To reduce the risk of recurrence and postoperative anal incontinence, external sphincteroplasty was performed. The patient underwent

monitoring for 8 weeks to evaluate wound healing and detect any post-operative complications. Fortunately, it was observed that he recovered smoothly without any significant complications. Though fistulas typically afflict individuals aged 30-70 years¹³ the case of a 23-year-old male patient presenting with a complex fistula is uncommon and warrants further investigation.

CONCLUSION:

This study highlights a 23-year-old patient diagnosed with a complex and unusual case of fistula-in-ano, characterized by three external openings and the absence of an internal opening. Managing complex fistulas is challenging due to the increased risks of anal incontinence and recurrence. In this particular case, fistulectomy and laser ablation were performed to mitigate the likelihood of recurrence. Additionally, immediate sphincteroplasty was undertaken to minimize the risk of anal incontinence. Subsequent follow-ups revealed the patient to be in good health with no reported complaints.

Main Novel Aspects:

This patient report presents a rare case of an anal fistula with three external openings.

MR Fistulography imaging was not able to detect any internal opening. To treat the patient and prevent any future complications, laser ablation along with sphincteroplasty was performed.

Declarations:

- Funding:** This research received no specific grant from any funding agency
- Conflict of interest:** The authors declare that they have no conflict of interest.
- Ethics approval:** This study did not require any ethical approval from the institutional review board (IRB). The patient was informed, and permission was obtained to use the medical data.
- Consent to participate:** The patient was informed, and permission was obtained to use the medical data.
- Consent for publication:** Written consent was obtained for publication.
- Availability of data and material:** Not applicable
- Code availability:** Not applicable.
- Authors' contributions:**

PN: Conception, drafting, final approval. MAA: Conception and cordially made the treatment decisions. VK: Critically revising. MMH: Adding important intellectual content. All authors commented on previous versions of the manuscript and read and approved the final manuscript.

REFERENCES:

- Andreou C, Zeindler J, Oertli D, Misteli H. Longterm outcome of anal fistula-a retrospective study. *Sci Rep.* 2020;10(1):6483.
- Babu AK, Naik MB, Babu MR, Madhulikia M. Seton-as a gold standard treatment for high fistula in ano. *J. evid. based med. healthc.* 2015;2(11):1687-1693.
- Gupta PJ. Radiofrequency fistulotomy in anal fistula. An alternative to conventional surgical fistulotomy. *Medicina (Kaunas).* 2003;39(10):996-998.
- Sheikh P. Controversies in fistula in ano. *Indian J Surg.* 2012;74(3):217-220.
- Hong Y, Qiu Y, Li G. A case report of primary complex anal fistula with 7 external openings treated with combined preoperative 3D MRI model. *Medicine (Baltimore).* 2023;102(11):e33264.
- Gupta PJ. A case of multiple (eight external openings) tubercular anal fistulae. *Case report. Eur Rev Med Pharmacol Sci.* 2007;11(5):359-361
- Champagne BJ, O'Connor LM, Ferguson M, Orangio GR, Schertzer ME, Armstrong DN. Efficacy of anal fistula plug in the closure of cryptoglandular fistulas: long-term follow-up. *Dis Colon Rectum.* 2006;49(12):1817-1821.
- Rojanasakul A, Pattanaarun J, Sahakitrungruang C, Tantiphlachiva K. Total anal sphincter saving technique for fistula-in-ano; the ligation of intersphincteric fistula tract. *J Med Assoc Thai.* 2007;90(3):581-586.
- Cheung FY, Appleton ND, Rout S, Kalaiselvan R, Nicholson JA, Samad A, Chadwick M, Rajaganesan R. Video-assisted anal fistula treatment: a high-volume unit initial experience. *Ann R Coll Surg Engl.* 2018;100(1):37-41.
- Hong Y, Qiu Y, Li G. A case report of primary complex anal fistula with 7 external openings treated with combined preoperative 3D MRI model. *Medicine (Baltimore).* 2023;102(11):e33264
- Woo K, Ulloa J, Allon M, Carsten CG 3rd, Chemla ES, Henry ML, Huber TS, Lawson JH, Lok CE, Peden EK, Scher L, Sidawy A, Maggard-Gibbons M, Cull D. Establishing patient-specific criteria for selecting the optimal upper extremity vascular access procedure. *J Vasc Surg.* 2017;65(4):1089-1103.e1
- Anaraki F, Etamad O, Abdi E, Bagherzadeh G, Behboo R. Assessment of fistulectomy combined with sphincteroplasty in the treatment of complicated anal fistula. *J. Coloproctol.* 2017; 37:232-237.
- Sanchez-Haro E, Vela E, Cleries M, Vela S, Tapiolas I, Troya J, Julian JF, Parés D. Clinical characterization of patients with anal fistula during follow-up of anorectal abscess: a large population-based study. *Tech Coloproctol.* 2023;27(10):897-907.