



Gastrointestinal Imaging Case Report

Perianal Inflammation or Something More?

Romina Geraldine Dsouza¹, Mohamed Rizwan Haroon Al Rasheed², Gavin Low¹

¹Departments of Radiology and Diagnostic Imaging and ²Pathology, University of Alberta, Edmonton, Alberta, Canada.



***Corresponding author:**

Romina Geraldine Dsouza,
Department of Radiology and
Diagnostic Imaging, University
of Alberta, Edmonton, Alberta,
Canada.

dsouza1@ualberta.ca

Received : 11 March 2021
Accepted : 06 August 2021
Published : 01 September 2021

DOI
10.25259/JCIS_54_2021

Quick Response Code:



ABSTRACT

Perianal fistulae are commonly complicated by abscesses and ramifications, which have well-recognized imaging morphology. Less commonly, atypical findings of solid enhancing mass-like lesions with no fluid component are associated with chronic and recurrent fistulae, the etiology of which includes inflammatory masses as well as locally aggressive malignancy. The latter predicts poorer prognosis and warrants extensive surgical resection. The reading radiologist must identify the unusual appearance, be aware of the possible etiologies, and if appropriate recommend prompt tissue sampling to exclude malignancy. This is pertinent as it determines surgical management, which is crucial in achieving a potential curative outcome.

Keywords: Perianal fistula, Perianal mucinous adenocarcinoma, Carcinoma in fistula in ano, Perianal phlegmon

INTRODUCTION

Perianal fistulous disease is the cause for a large percentage of colorectal morbidity in North America. Before the year 2000, imaging played a limited role in the treatment of this condition. Subsequently, however, magnetic resonance imaging (MRI) has established itself as being a pivotal pretreatment requirement, its most important contribution being to delineate fistula anatomy and its complex and sometimes clinically hidden extensions. In the case presented here, one such complication, a uniformly enhancing ischioanal nodule along the trans-sphincteric fistula tract, was identified. Due to its unconventional imaging appearance, it was considered unlikely to be a simple abscess, the usual suspect in fistula in ano. An imaging differential diagnosis of mucinous adenocarcinoma, a rare but known complication of this disease versus an inflammatory phlegmon, was considered. Percutaneous biopsy was performed confirming inflammation with the absence of malignancy. MRI with histopathological correlation thus directed effective surgical management.

CASE REPORT

A 52-year-old gentleman initially presented to our institution with a perianal fistula and abscess in June 2020. The abscess drained spontaneously and he responded to conservative management. Four months later, he returned with perianal pain and suspected recurrent abscess in the left gluteal region. He had no pyrexia or systemic symptoms. White blood cell count was normal at $5.4 \times 10.9/L$, hemoglobin was also normal at 145 g/L. Pelvic MRI was performed which delineated a left trans-sphincteric fistula leading to a T2 hyperintense ovoid uniformly hyperenhancing focus measuring approximately $2 \times 1.5 \times 3$ cm in the left ischioanal fossa [Figures 1 and 2]. The lesion showed no appreciable fluid component resulting in two main etiological considerations, first,

This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-Share Alike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

©2021 Published by Scientific Scholar on behalf of Journal of Clinical Imaging Science

a neoplasm arising from a longstanding perianal fistula and second, a solid inflammatory phlegmon.

An ultrasound-guided biopsy was performed, [Figure 3] in view of possible malignancy, which showed suppurative inflammation and granulation tissue with no evidence of malignancy [Figure 4]. The surgical team, therefore, went ahead with a simple excision of the lesion and fistula tract. Final excision specimen confirmed an inflammatory phlegmon with dense acute inflammatory infiltrate forming microabscess as well as granulation tissue with reactive endothelial cells. A degenerated hair follicle was also noted at the center of the inflammatory focus. Absence of malignancy was confirmed by pathology.

DISCUSSION

A perianal fistula is defined as a non-anatomic communication between the anal or anorectal mucosa and the perianal skin. Most often being secondary to anal cryptoglandular infection and abscess, complex high fistulae without a definite anal canal opening cannot be adequately explained by this hypothesis. Fistula in ano is documented to be associated with fistulizing diseases including 25% of patients being followed up for Crohn's disease,^[1] active diverticulitis, tuberculosis, and other pelvic infection. It also complicates trauma, vaginal delivery, anorectal malignancy, and locoregional radiation.^[2]

Studies quote the prevalence of perianal fistulae as being 0.01% predominantly in the young adult.^[1] These patients present with severe pain, especially during stool passage, perianal excoriation with local swelling, or even obvious discharge. Despite being a benign, inflammatory process, it is not without immense patient discomfort and tremendous, sometimes extended morbidity. Preliminary treatment includes abscess drainage, interval seton placement, and, if persistent, excision of the fistula tract. The primary cause of the protracted course of the illness and high incidence of recurrence is the residual and/or untreated fistula extensions. Herein lies the role of MRI, the imaging modality of choice.^[2]

On MRI, an active perianal fistula is visualized as a linear high signal intensity tract on fat saturated T2-weighted images with peripheral enhancement on post-gadolinium T1-weighted fat-saturated sequences. An associated abscess, usually occurring in an active fistula, is characteristically T1 hypointense, T2 mildly hyperintense with the central purulent fluid component demonstrating diffusion restriction, and no internal contrast enhancement. The surrounding granulation tissue wall enhances with the classic peripheral/rim enhancement.^[1]

Less commonly, like in our case, instead of the well-known imaging appearance of an abscess, one may encounter T2 hyperintense focal abnormalities associated with the fistula

tracts that demonstrate uniform mass-like enhancement with no discernable fluid component. This rules out a liquefied abscess.

It now becomes important to consider a true mass as well as a pseudomass and not assume the T2 hyperintense focus represents the much more frequently encountered infective abscess. This is especially true in chronic fistulizing conditions like Crohn's disease.^[3] In these circumstances, clinical presentation is not significantly different from the recurrent fistula itself, and MRI usually gives the first indication of a possibly concerning underlying lesion.

MRI features of pre-contrast T1 hypointensity and T2 hyperintensity alone are impossible in differentiating a malignancy from an abscess. It is thus imperative for a radiologist to carefully consider a few subtle clues to help make a more accurate diagnosis or at least raise the possibility for concern when relevant. The presence of heterogeneous, intensely high T2 signal that demonstrates uniform, and solid mass-like enhancement must increase one's index of suspicion. The absence of a non-enhancing fluid collection and relatively less surrounding tissue edema and stranding also makes a mass or pseudo mass a likely possibility.^[4] The presence of enhancing soft tissue expanding the fistula also suggests possible malignant transformation.^[5]

As malignancy arising in fistula in ano is rare with only a few reports in the literature, no definite diagnostic criteria have been approved. Rosser *et al.*^[6] first described the association in 1934. The proposed diagnostic criteria for this entity include the fistula having to antedate the carcinoma by a minimum of 10 years, the tumor being the only tumor in the ano-rectum and the internal opening of the fistula having to specifically open into the anal canal and not into the lesion alone.^[6]

Anal canal malignancies are classified into anal canal tumors which are predominantly epithelial in origin, and anal margin malignancies, which include malignancies arising from chronic fistulae.^[4] Most commonly mucinous adenocarcinoma and much less commonly squamous cell carcinoma and basal cell carcinoma have been documented to arise in a chronic perianal fistula.^[6] Exact etiology of mucinous adenocarcinomas is still unknown. Degenerating fistula with associated mucosal regeneration is one consideration, the other being seeding of the fistula with neoplastic cells from higher up in the gastrointestinal tract.^[7]

Mucinous adenocarcinomas are not metabolically active on fluorodeoxyglucose positron emission tomography making histopathology the only definitive diagnostic tool.^[7] The presence of extracellular mucin lakes contributing to the high T2 signal with adjacent well-differentiated dilated glands, confirms the diagnosis.^[8] Being locally aggressive with high rates of local recurrence, abdominoperineal resection is the preferred treatment option with or without neoadjuvant

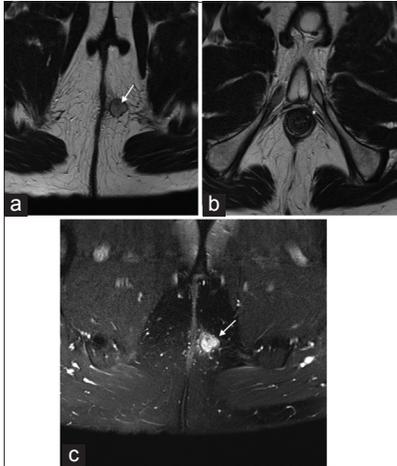


Figure 1: A 52-year-old male patient with a history of perianal fistula previously managed conservatively represents with perianal pain. (a) Unenhanced T2-weighted axial MRI showing a 2 cm well-circumscribed T2 hyperintense mass-like lesion (white arrow) in the left ischioanal fossa with a thin hypointense wall and minimal surrounding spiculation. (b) T2-weighted axial MRI at the level of the lower rectum showing the internal opening (asterisks) of the fistula in ano at 2' o clock position. (c) T2-weighted fat-saturated axial MRI image demonstrating the true high signal of the pseudo mass (white arrow).

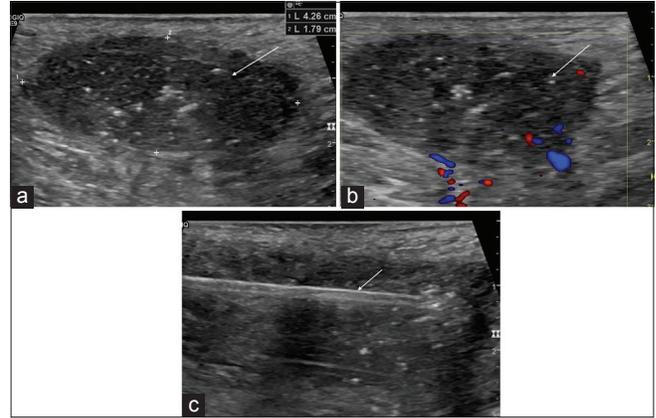


Figure 3: Our 52-year-old male patient underwent pre-surgical perineal ultrasound guided biopsy: (a) High-frequency B mode ultrasound image using a linear 6–15 Hz transducer showing the lesion on MRI to correspond to a well-circumscribed heterogeneously hypoechoic lesion with echogenic foci within (white arrow). (b) Color Doppler image of the same lesion demonstrating mild surrounding vascularity with no internal vascularity (white arrow). (c) B mode ultrasound confirming the 16 G biopsy needle (white arrow) within the lesion during ultrasound-guided tissue sampling for pre-operative tissue diagnosis.

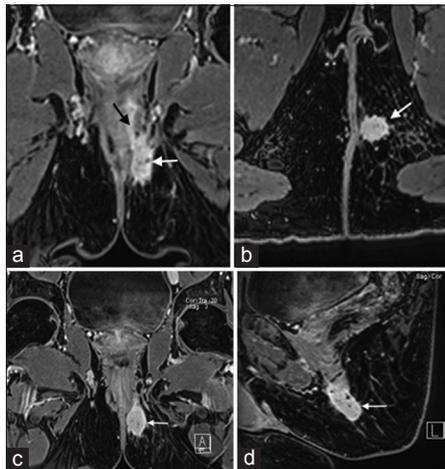


Figure 2: The same 52-year-old gentleman at the time of second presentation. (a) Post-contrast T1-weighted MRI in coronal orientation clearly delineating the enhancing left transphincteric fistula tract (black arrow) with uniform mass-like enhancement of the adjacent left ischio anal lesion (white arrow). Post-contrast (b) axial, (c) coronal, and (d) sagittal T1-weighted images confirm near-complete uniform enhancement of the lesion (white arrow).

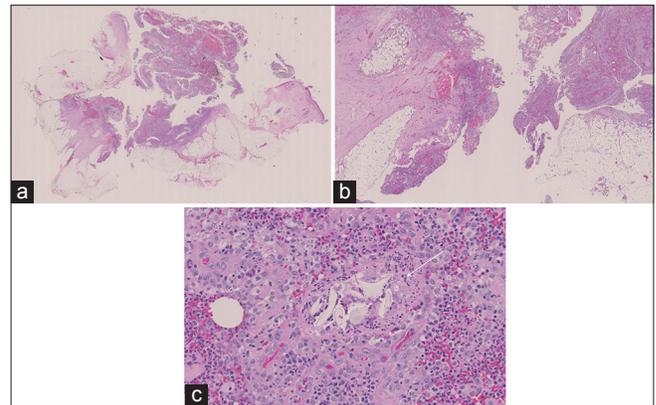


Figure 4: The 52-year-old gentleman then underwent surgical excision of the lesion. (a) Low-power image, H and E, original magnification $\times 4$, showing fragments of skin and subcutaneous adipose tissue with a disrupted fistula tract. (b) H and E, original magnification $\times 20$, demonstrating the fistulous tract within fibroadipose tissue with marked acute inflammation and fibrosis. (c) High-power image, H and E, original magnification $\times 200$, revealing dense acute inflammatory infiltrate forming microabscess as well as granulation tissue with reactive endothelial cells. A degenerated hair follicle is also noted at the center (white arrow).

chemo radiation. Distant metastasis is uncommon though lymphatic dissemination to inguinal nodes has been documented.^[7]

Solid enhancing perianal lesions are also seen in perianal Paget's disease, Bowens disease, as well as verrucous

perianal carcinoma, however, the cutaneous manifestations of these conditions result in early diagnosis.^[5] The relative T2 hyperintensity of an aggressive angiomyxomas makes it necessary to differentiate it from a mucinous adenocarcinoma described above. This rare benign tumor occurs in middle age women and usually involves the perianal tissue with

extension into multiple compartments. A characteristic laminated appearance on T2 and no associated fistula aids its identification.^[9] Other solitary perianal malignancies include a high-grade sarcoma and mesenchymal tumors such as solitary fibrous tumor and myofibroma. The latter two usually demonstrate lower T2 signal secondary to their fibrous content but malignant degeneration can increase the T2 signal intensity.^[5] These lesions may show hyperenhancement post-contrast but usually are not associated with fistula in ano. Once again, final diagnosis demands pathology correlation.

The atypical occurrence in a perianal fistula is what we ultimately found in our case where the enhancing mass-like soft tissue proved to be an inflammatory pseudo mass of dense acute inflammatory infiltrate with microabscess and granulation tissue. Although rarely detailed in literature, this can occur in chronic/recurrent fistulae similar to other locations in the body, secondary to long-standing low-grade infection.^[3] Hidradenitis suppurative, a condition arising secondary to occlusion of perianal follicular ducts with resultant duct rupture, inflammation, and fibrosis can mimic this presentation. MRI appearance similar to our case with or without abscess formation is typical. Although a degenerated hair follicle was noted within the inflammatory mass in our case as well, the presence of the fistulous opening into the anal canal differentiates a complicated fistula in ano from hidradenitis where no such communication is found.^[10] These conditions require simple excision, however, the surgical decision is possible only after exclusion of more ominous etiologies as described above.

CONCLUSION

The finding of a solid enhancing lesion in a chronic or recurrent perianal fistula demands a high index of suspicion for pathologies, especially mucinous malignancies that are known to arise within such fistulae. The radiologist reading the MRI must be aware of the differential diagnosis of such a finding, thus alerting the unsuspecting clinician and directing prompt histopathological evaluation. Treatment decisions and patient prognosis are greatly influenced by timely diagnosis and appropriate management.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. de Miguel Criado J, del Salto LG, Rivas PF, del Hoyo LF, Velasco LG, de las Vacas MI, *et al.* MR imaging evaluation of perianal fistulas: Spectrum of imaging features. *Radiographics* 2011;32:175-94.
2. O'Malley RB, Al-Hawary MM, Kaza RK, Wasnik AP, Liu PS, Hussain HK. Rectal imaging: Part 2, Perianal fistula evaluation on pelvic MRI-what the radiologist needs to know. *AJR Am J Roentgenol* 2012;199:W43-53.
3. Alvarez-Laso CJ, Moral S, Rodríguez D, Carrocera A, Azcano E, Cabrera A, *et al.* Mucinous adenocarcinoma on perianal fistula. A rising entity? *Clin Transl Oncol* 2018;20:666-9.
4. Cicero G, Ascenti G, Blandino A, Pallio S, Abate C, D'Angelo T, *et al.* Magnetic resonance imaging of the anal region: Clinical applications. *J Clin Imaging Sci* 2020;10:76.
5. Surabhi VR, Menias CO, Amer AM, Elshikh M, Katabathina VS, Hara AK, *et al.* Tumors and tumorlike conditions of the anal canal and perianal region: MR imaging findings. *Radiographics* 2016;36:1339-53.
6. Yang BL, Shao WJ, Sun GD, Chen YQ, Huang JC. Perianal mucinous adenocarcinoma arising from chronic anorectal fistulae: A review from single institution. *Int J Colorectal Dis* 2009;24:1001-6.
7. Díaz-Vico T, Fernández-Martínez D, García-Gutiérrez C, Suárez-Sánchez A, Cifrián-Canales I, Mendoza-Pacas GE, *et al.* Mucinous adenocarcinoma arising from chronic perianal fistula-a multidisciplinary approach. *J Gastrointest Oncol* 2019;10:589-96.
8. Baars JE, Kuipers EJ, Dijkstra G, Hommes DW, de Jong DJ, Stokkers PC, *et al.* Malignant transformation of perianal and enterocutaneous fistulas is rare: Results of 17 years of follow-up from The Netherlands. *Scand J Gastroenterol* 2011;46:319-25.
9. Surabhi VR, Garg N, Frumovitz M, Bhosale P, Prasad SR, Meis JM. Aggressive angiomyxomas: A comprehensive imaging review with clinical and histopathologic correlation. *AJR Am J Roentgenol* 2014;202:1171-8.
10. Balci S, Onur MR, Karaosmanoğlu AD, Karçaaltıncaba M, Akata D, Konan A, *et al.* MRI evaluation of anal and perianal diseases. *Diagn Interv Radiol* 2019;25:21-7.

How to cite this article: Dsouza RG, Al Rasheed MR, Low G. Perianal inflammation or something more? *J Clin Imaging Sci* 2021;11:47.