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Fistula-In-Ano (*Bhangandara*) With Retroperitoneal Extension Forming Retroperitoneal Abscess

A CASE REPORT

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INTRODUCTION

'*Bhagandara*' is the disease, which causes splitting or discontinuity in the region of *'Bhaga'*, *'Vasti'* (Perineal)and '*Guda*' (Perianal) region. In Ayurveda, *Bhagandara* has been mentioned as one among *Ashtamaharoga* (eight major diseases) i.e. the disease that is difficult to cure because of its callous attitude[1]. A fistula that goes untreated generally won't heal on its own. This can lead to long-term complications, such as: Persistent infection, Fistula extension, Cancer[2]. The disease in which *Bhaga*, *Guda* and *Basti Pradesha* becomes *Vidaarita* (get torn) is known as *Bhagandara*. Here we encountered with such a case where the *samprapti* has gone beyond the *Bhaga pradesha*. Fistula in ano is the entity which is correlated with *Bhagandara*.

Though fistula-in-ano and anorectal abscesses are commonly encountered in surgical departments, but their extension into the retroperitoneum and pelvis to form an extensive collection is rare. In this article the case of a 38-year-old male who diagnosed for retroperitoneal collection, operated for the same has been described. The patient's condition improved with further treatment and local wound care, and he was subsequently discharged.

Retroperitoneal abscesses resulting from perianal abscesses are infrequent in surgical practice. Unlike the intraperitoneal region, the retroperitoneum typically responds very less to bacterial contamination. As a result, retroperitoneal abscesses often advance silently, exhibiting minimal symptoms and progressing

slowly over time. This chronic and asymptomatic course presents significant challenges in both diagnosis and management. Delayed diagnosis and inadequate drainage can markedly elevate the risk of mortality and sepsis. Despite treatment endeavors, mortality rates range from 11% to 20% among patients diagnosed with this condition [3].

CASE REPORT-

Patient information-

A 38-year-old man arrived at the surgery OPD with complaints of lower abdominal pain, fever along with pain and discharge from anal region. He exhibited signs of sepsis, including tachycardia, hypotension, and tachypnea since 2-3 days. Patient had surgical history of partial fistulectomy with drain insertion in supralevator space 2 months ago.

Figure 1,2- MRI report before previous surgey

MR FISTULOGRAM (PLAIN)

Clinical profile : Pus discharge in perianal region.

Technique : Multiplanar, multiecho MR of the fistulogram was performed. Marker has been kept at the external opening.

Findings :
A complex fistulous tract is seen on left side with secondary extensions of the tract (described below). The primary tract is ~ 9 cm in length and 6 mm in width. It appears hypointense on T1 and T2W images and hyperintense on STIR images. It has thick hypointense margins on T2W images. Mild hyperintense signal noted within the surrounding fat on STIR images suggestive of edema. Mild surrounding fat stranding also noted.

A suspicious internal opening of the tract is seen in anal canal at 5-6 o'clock location at 4.5 cm above the anal verge. From internal opening, it is coursing inferiorly and posteriorly with external opening in left gluteal region.

It shows a horse-shoe shaped secondary tract in posterior perinanal region, at 4.7 cm above the level of anal verge.

It shows another secondary tract with suprasphincteric extension. It is penetrating the left levator ani at 4-5 o'clock location with internal opening in distal rectum at 4-5 o'clock location, at 8 cm above the anal verge.

A small well defined focal collection measuring 1 x 1.5 x 2.4 cm is seen in anterior wall of distal rectum, (just above levator ani) in submucosal plane.

Multiple prominent reactive pararectal lymph nodes seen measuring up to 11 x 6 mm.

There is no involvement of major pelvic organs.
Ischio-rectal and ischio-anal fossae appear normal bilaterally.
Rest of the visualised pelvic structures do not show any significant abnormality.
Visualised bones do not show any marrow signal abnormality.

IMPRESSION :

- MR Fistulogram reveals a complex fistulous tract on left side with secondary extensions of the tract (as described) with suspicious internal opening of the tract in anal canal at 5-6 o'clock location at 4.5 cm above the anal verge. From internal opening, it is coursing inferiorly and posteriorly with external opening in left gluteal region.
- It shows a horse-shoe shaped secondary tract in posterior perinanal region, at 4.7 cm above the level of anal verge.
- It shows another secondary tract with suprasphincteric extensio, penetrating the left levator ani at 4-5 o'clock location with internal opening in distal rectum at 4-5 o'clock location, at 8 cm above the anal verge.
- A small well defined focal collection in anterior wall of distal rectum, (just above levator ani) in submucosal plane.
- Multiple prominent reactive pararectal lymph nodes

Figure 3- MRI film showing complex fistula(before previous surgery)

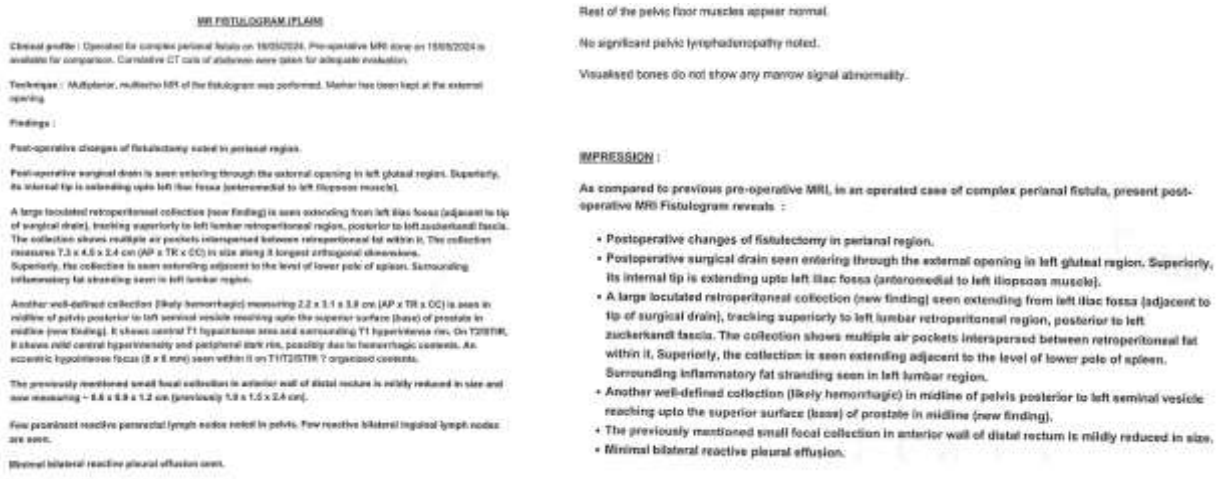
Clinical examination

Clinical examination revealed tenderness in the lower abdomen, and a fistula was noted during a rectal examination. The patient reported intermittent pustular discharge from the perianal region for the past 2 months.

Diagnostic assessment

An ultrasound of the abdomen and pelvis done which showed heterogeneous fluid collections in pelvis on left side & extending into the left iliac fossa. Considering the 2 months history of a fistula, the patient was investigated with an MRI fistulogram which revealed localized retroperitoneal collection extending from left iliac fossa to left lumbar retroperitoneal region measures 7.3 cm x 4.5 cm x 2.4 cm. Another collection measuring 2.2 cm x 3.1 cm x 3.0 cm in midline of pelvis posterior to left seminal vesicle. The blood picture showed anemia and leucocytosis.

Figure 4,5 – MRI report before second surgery



MR FISTULOGRAM (PLAIN)

Clinical profile : Operated for complex perianal fistula on 18/03/2024. Pre-operative MRI done on 15/05/2024 is available for comparison. Contrastive CT cuts of abdomen were taken for adequate evaluation.

Technique : Multiplanar, multiecho MR of the fistulogram was performed. Marker has been kept at the external opening.

Findings :

Post-operative changes of fistulectomy noted in perianal region.

Post-operative surgical drain is seen entering through the external opening in left gluteal region. Superiorly, its internal tip is extending upto left iliac fossa (anteromedial to left iliopsoas muscle).

A large loculated retroperitoneal collection (new finding) is seen extending from left iliac fossa (adjacent to tip of surgical drain), tracking superiorly to left lumbar retroperitoneal region, posterior to left zuckerkandl fascia. The collection shows multiple air pockets interspersed between retroperitoneal fat within it. The collection measures 7.3 x 4.5 x 2.4 cm (AP x TR x CC) in size along 3 longest orthogonal dimensions. Superiorly, the collection is seen extending adjacent to the level of lower pole of spleen. Surrounding inflammatory fat stranding seen in left lumbar region.

Another well-defined collection (likely hemorrhagic) measuring 2.2 x 3.1 x 3.8 cm (AP x TR x CC) is seen in midline of pelvis posterior to left seminal vesicle reaching upto the superior surface (base) of prostate in midline (new finding). It shows central T1 hypointense area and surrounding T1 hyperintense rim. On T2/STIR, it shows mild central hyperintensity and peripheral dark rim, possibly due to hemorrhagic contents. An eccentric hypointense focus (8 x 6 mm) seen within it on T1/T2/STIR ? organized contents.

The previously mentioned small focal collection in anterior wall of distal rectum is mildly reduced in size and now measuring ~ 8.6 x 6.9 x 1.2 cm (previously 1.8 x 1.5 x 2.4 cm).

Few prominent reactive pararectal lymph nodes noted in pelvis. Few reactive bilateral inguinal lymph nodes are seen.

Minimal bilateral reactive pleural effusion seen.

Rest of the pelvic floor muscles appear normal.

No significant pelvic lymphadenopathy noted.

Visualised bones do not show any marrow signal abnormality.

IMPRESSION :

As compared to previous pre-operative MRI, in an operated case of complex perianal fistula, present post-operative MRI Fistulogram reveals :

- Postoperative changes of fistulectomy in perianal region.
- Postoperative surgical drain seen entering through the external opening in left gluteal region. Superiorly, its internal tip is extending upto left iliac fossa (anteromedial to left iliopsoas muscle).
- A large loculated retroperitoneal collection (new finding) seen extending from left iliac fossa (adjacent to tip of surgical drain), tracking superiorly to left lumbar retroperitoneal region, posterior to left zuckerkandl fascia. The collection shows multiple air pockets interspersed between retroperitoneal fat within it. Superiorly, the collection is seen extending adjacent to the level of lower pole of spleen. Surrounding inflammatory fat stranding seen in left lumbar region.
- Another well-defined collection (likely hemorrhagic) in midline of pelvis posterior to left seminal vesicle reaching upto the superior surface (base) of prostate in midline (new finding).
- The previously mentioned small focal collection in anterior wall of distal rectum is mildly reduced in size.
- Minimal bilateral reactive pleural effusion.

Surgical intervention

The fistulous tract was ligated at intersphincteric space in lithotomy position and the localized collection was drained from the supralevator space.

An exploration of retro peritoneum was performed. Pfannenstiel incision made. Collection drained from retroperitoneal approach from left side and Romovac drain no. 18 kept in left iliac and pelvis of retroperitoneal region.

Following surgery, the patient's condition showed significant improvement, and he was treated with broad-spectrum antibiotics. Culture-specific antibiotics were identified and administered accordingly. The wound received local care under aseptic precautions for one month, and the Romovac drain in the retroperitoneal space was removed after a repeat ultrasound confirmed no residual collection. The patient's general condition improved, and he was discharged with instructions for regular dressings and follow-up appointments.

Table 1: Timeline of events

Timeline	Event	Details
Two months prior	Initial symptoms –Purulant discharge through postule over gluteal region	Intermittent pustular discharge and pain in the perianal regionwere noticed.
	Partial fistulectomy	Partial fistulectomy was performed, and a drain was inserted in the supralevator space.
Post-surgical period	Persistent symptoms	Pain, fever and lower abdominal discomfort were experienced for 2 months.

At admission	Persisting symptoms, Pain in abdomen	Lower abdominal pain, fever and perianal discharge were reported.
	Clinical signs	Sepsis was observed; tachycardia, hypotension, and tachypnea were noted.
Initial investigations	Ultrasound	Heterogeneous fluid collection in the pelvis and left iliac fossa were revealed.
	MRI Fistulogram	Retroperitoneal collections were identified.
Surgical intervention	Procedure	Exploration of retro peritoneum was performed via Pfannenstiel incision.
	Findings	Retroperitoneal collections were drained, and the supralevator fistulous tract was ligated.
	Drain placement	A romovac drain was placed in the left iliac region and retroperitoneal space.
Post-surgical recovery	Treatment	Broad spectrum antibiotics were initiated and later adjusted to culture specific antibiotics.
	Wound care	Local wound care was provided under aseptic precautions for one month.
	Imaging	Repeat ultrasound showed no residual collections, and the drain was removed.
Discharge and follow-up	Discharge	The patient was discharged with instructions for regular dressings and follow-ups.
	Outcome	Significant improvement in general condition was noted at discharge.

Discussion

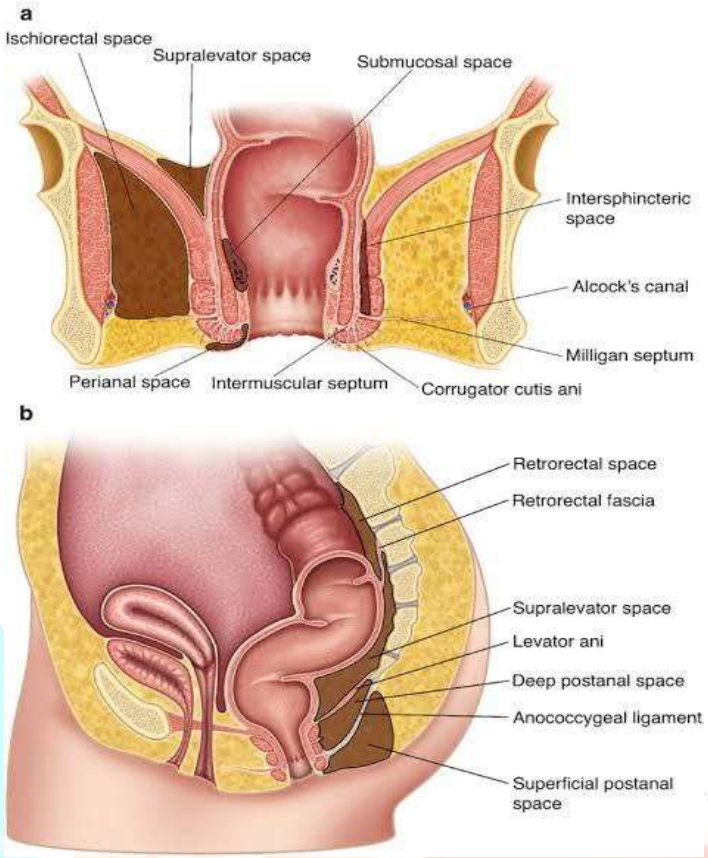
Anorectal abscesses are a common surgical concern encountered in everyday medical practice, primarily arising from non-specific cryptoglandular infections. Depending on their site, anorectal abscesses are categorized into perianal, ischiorectal, intersphincteric, and supralevator types. Ayurveda explained *Guda vidradhi* (Anal abscess) under the classification of *Abhyantara-Vidradhi*, which occurs at *Guda pradesh* (anal region). It's repair and treatment by means of *Saptoupkrama* (seven types of treatment) is the fundamental major contribution of Acharya Sushruta[4]. *Guda vidradhi* co-related with anal abscess on the basis of symptom.

Formation of retroperitoneal abscess in case of Fistula in Ano is uncomman and Multiple factors may contribute in this. These include perforations caused by colorectal cancers, diverticulitis, retroperitoneal appendicitis, pancreatitis, pancreatic cancer, inflammatory bowel diseases, urinary tract obstruction, osteomyelitis, perforations from postoperative duodenal ulcers, pelvic and postpartum infections, trauma, and the dissemination of infection via the bloodstream or lymphatic system from distant sites. Conditions such as diabetes mellitus, chronic alcohol consumption, glucocorticoid use, malignancies, and distant infections can elevate the risk of complications by impairing the host immune response. In our case, we encountered a rare scenario of the existence of the connection between the fistula-in-ano and the multiple retroperitoneal suppurative collections which was a diagnostic, clinical, and surgical challenge[3].

Perianal abscesses constitute about 60% of all anorectal abscesses[5]. They typically originate from infections in the perianal glands within the intersphincteric space and can extend upwards to form a supralevator abscess. Symptoms commonly involve perianal pain and fever. Patients with diabetes or compromised immune systems are particularly vulnerable. Complications associated with anorectal abscesses include external rupture through the perianal skin or internal rupture into the anal canal, potentially leading to the formation of fistula-in-ano. However in immunocompromised patients, clinical presentation may deviate from the typical pattern, with abscesses occurring in atypical anatomical locations[6].

Without prompt diagnosis and appropriate treatment, it is expected that more than 90% of these collections will rupture[7]. The puborectalis sling exerts significant pressure on the posterior rectal wall at the anorectal junction, typically preventing abscess extension above this level into the supralevator spaces. Instead, abscesses often rupture through the longitudinal muscle and spread trans-sphincterically into one of the infralevator anorectal spaces[8].

Figure 6– showing supralevator space and anal sphincters

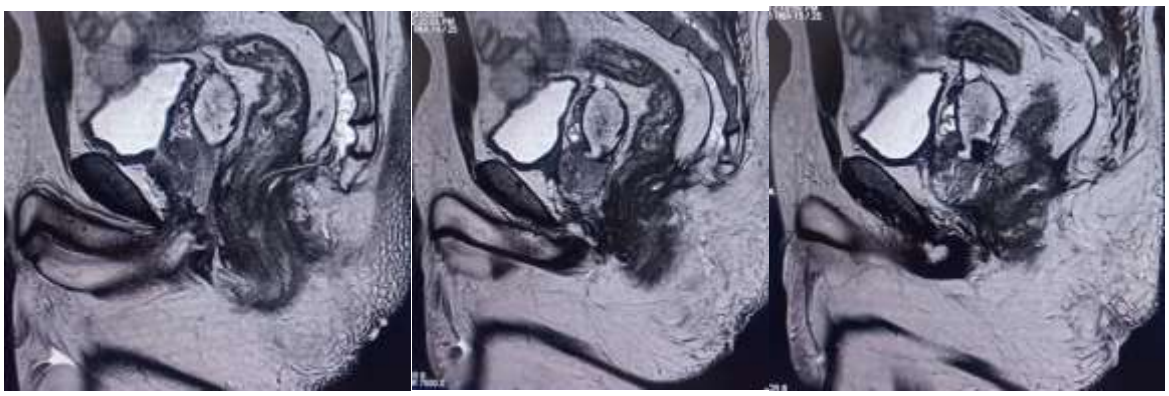


However, in rare instances, a low intersphincteric abscess may extend above the puborectalis muscle to form a high intersphincteric abscess, which can rupture into the supralevator spaces[9]. Supralevator abscesses can spread into the prevesicle space due to direct communication from the pararectal space, where the umbilicovesicle fascia terminates at the reflection of the vesicle peritoneum. From the prevesicle space, infection may disseminate anteriorly throughout the Retzius space and into other pelvic compartments, or posteriorly into the retroperitoneum through direct spread[10]. The retroperitoneum typically responds less vigorously to bacterial contamination compared to the intraperitoneal region, often leading to a more subtle and asymptomatic course. This may result in delayed detection and treatment, potentially increasing the risk of sepsis and mortality.

A clinician can only partially assess the retroperitoneum through physical examination, and laboratory tests provide limited information. Therefore, radiological investigations are crucial for accurate diagnosis. MRI has demonstrated effectiveness in assessing the relationship of complex horseshoe abscesses with the deep postanal region, as well as in evaluating secondary abscess formations, additional fistulas, internal openings, the extent of abscess spread, and sphincter involvement[11]. In this case, MRI played a vital role in diagnosis. Deep-spreading perianal abscesses are considered severe in anorectal disease due to their rare occurrence and subtle clinical symptoms, which may lead to delayed diagnosis, severe sepsis, and potentially fatal outcomes[12]. The absence of typical signs alongside abdominal pain can complicate diagnosis for physicians evaluating abdominal pathology. Retroperitoneal abscesses can stem from the genitourinary tract, be idiopathic, postoperative, or occasionally involve other organs such as the colon, duodenum, and pancreas. Typically, treatment involves lumbar incisions, as the transperitoneal approach is

less effective. The mortality rate can reach 26%, especially among critically ill patients with delayed diagnosis[13].

Figure 7,8,9- MRI film showing retroperitoneal extention of abscess



Treatment approaches are diverse, typically favoring open drainage and debridement as the established norm. Managing limited supralevator abscesses presents challenges in percutaneous intersphincteric drainage, where fluoroscopic monitoring may prove insufficient, potentially leading to prolonged sepsis[14]. When dealing with extensive pre- or retroperitoneal extension, it is advisable to avoid accessing the peritoneal cavity to reduce the risk of contamination and secondary peritonitis[15]. Effective treatments include drainage by abdominal incisions or performing extraperitoneal drainage through lower midline abdominal incisions, typically closed primarily with drains or vacuum-assisted devices[16].

In the past, surgery served as the mainstay treatment for retroperitoneal abscesses until the advent of imaging-guided percutaneous drainage offered an alternative. Initially used for uncomplicated abscesses, percutaneous drainage has advanced to handle complex cases involving multiple compartments or fistulas in regions such as the pancreas, spleen, or retroperitoneum. This minimally invasive approach has demonstrated significant advantages in lowering mortality and complications, particularly in high-risk patients or those with prior surgical history, compared to conventional surgical techniques[17].

Conclusions

This case report highlights the challenges encountered in surgical practice when dealing with rare perianal issues, emphasizing the complexities that challenge surgeons in various aspects. A thorough understanding of the perianal and retroperitoneal regions is crucial for surgeons to make informed decisions regarding surgical management. Surgical drainage of suprarectal abscesses is crucial in patient's care. A minimally invasive approach can have significant advantages over open abdominal surgery[18]. Radiodiagnosis plays a significant role both before and after surgery in managing such complex cases effectively.

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Declaration of patient consent

Authors certify that they have obtained a patient consent form, where the patient has given his consent for reporting the case along with the images and other clinical information in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

Conflicts of Interest- No any.

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