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Case Report

Surgery Section

Perianal Pilonidal Sinus Mimicking as Fistula-in-Ano: A Rare Diagnostic Dilemma

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ABSTRACT

Pilonidal sinus disease is a common condition that typically presents in the sacrococcygeal region of young adult males. However, its occurrence in the perianal region is exceedingly rare and often mimics more common anorectal pathologies such as fistula-in-ano or perianal abscess. This report describes a 17-year-old male who presented with a two-week history of perianal discharge, itching, and pain during defaecation. Clinical examination revealed an external opening at the 5 o'clock position with active pus discharge and raised sphincter tone. A Magnetic Resonance Imaging (MRI) fistulogram suggested a complex Grade V fistula-in-ano extending into the left supralevator region. Based on imaging and clinical findings, a diagnosis of fistula-in-ano was made. Intraoperatively, a fistulous tract containing hair and purulent material was discovered, raising suspicion for a pilonidal sinus. Histopathological examination confirmed the diagnosis, revealing abundant keratinous debris and hair shafts consistent with pilonidal disease. The patient underwent tract excision and recovered well with regular postoperative wound care. This case highlights the diagnostic challenge posed by perianal pilonidal sinus, which can closely resemble a complex anal fistula on both clinical and radiological grounds. Accurate diagnosis may only be possible intraoperatively and requires a high index of suspicion. Due to its rarity and potential for misdiagnosis, this case underscores the importance of considering pilonidal disease in the differential diagnosis of perianal fistulas, especially in young males, and contributes to the limited literature on this atypical presentation.

Keywords: Histology, Imaging, Inflammation, Perianal abscess

CASE REPORT

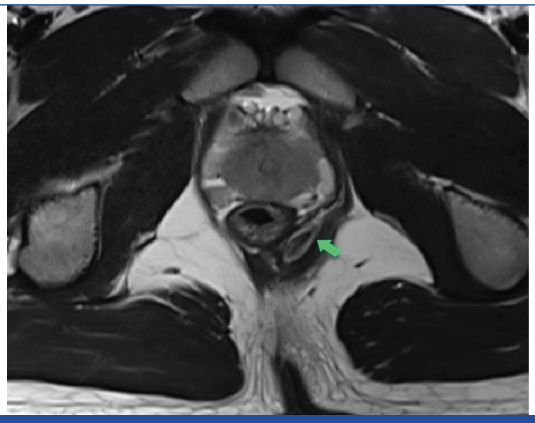
A 17-year-old male presented to the clinic with perianal discharge and itching for the past two weeks, associated with pain, especially during defaecation. There was no history of per rectal bleeding, fever, bowel irregularities, prolonged sitting, or trauma. The patient did not provide any prior history of similar complaints. He had an average body habitus and was not notably hirsute, making the presentation atypical for pilonidal sinus, which is commonly associated with prolonged sitting and excessive hair distribution.

On digital rectal examination, an external opening was identified at the 5 o'clock position with active purulent discharge. The anal sphincter tone was found to be raised, and the examining glove was stained with stool. There was no evidence of active per rectal bleeding. No fissure or internal fistulous opening was palpated.

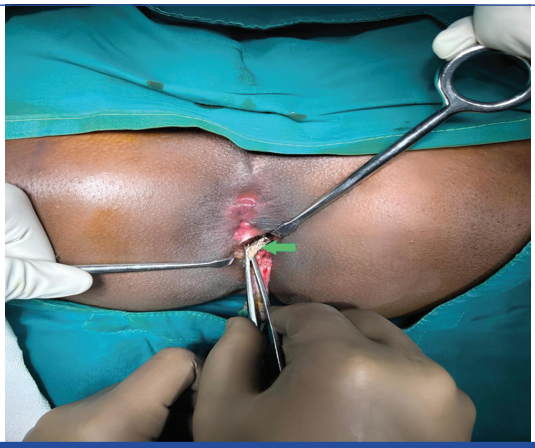
Based on the presence of a perianal external opening with ongoing discharge, associated pain during defaecation, raised sphincter tone, and the absence of systemic signs of infection, a high anal fistula was clinically suspected. The location and depth of the tract raised suspicion for a complex fistulous path, possibly extending into the supralevator space or ischioanal fossa. The differential diagnoses at this point included fistula-in-ano and perianal abscess.

The patient underwent a Magnetic Resonance Imaging (MRI) fistulogram, which showed a linear fistulous tract with an external opening at the 5 o'clock position, extending superiorly over an approximate length of 42 mm into the left supralevator region, ending in a well-defined collection approximately 21×21×19 mm in size. These findings were suggestive of a Grade V fistula in ano [1], with the external opening in the left perianal region (St James University Hospital Classification) [Table/Fig-1]. Consequently, a provisional diagnosis of fistula-in-ano was made.

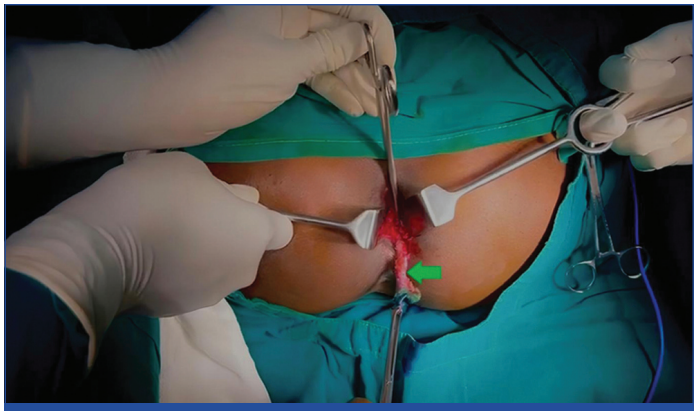
Based on the MRI findings and the clinical suspicion of a high anal fistula, the patient was scheduled for examination under anaesthesia and surgical exploration with fistula tract excision. Intraoperatively, a fistulous tract was noted, along with a collection of hair and pus discharge [Table/Fig-2]. The tract was excised, and the wound was left open to heal by secondary intention [Table/Fig-3]. The procedure was uneventful. The excised tract was sent for histopathological examination, which revealed abundant keratinous material and numerous hair shafts. A small fragment of tissue lined by keratinising stratified squamous epithelium was also observed. These features confirmed the diagnosis of a pilonidal sinus [Table/Fig-4].



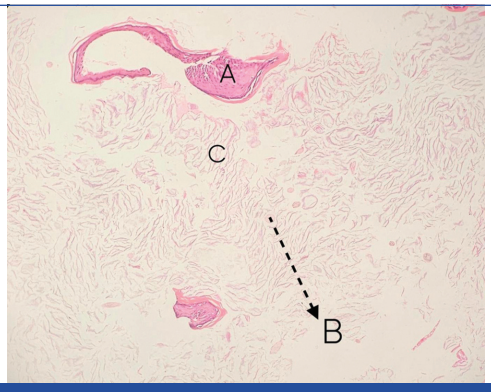
[Table/Fig-1]: Arrow showing fistulous tract opening in left supralevator region (T2-weighted MRI).



[Table/Fig-2]: Fistulous tract with collection of hair.



[Table/Fig-3]: Excision of entire tract.



[Table/Fig-4]: a) Small fragment of tissue lined by keratinising stratified squamous epithelium; b) Hair shafts; c) Abundant keratinous material (Haematoxylin and eosin staining; 40x).

Postoperatively, the patient underwent regular dressings twice daily with Betadine flush. The patient was discharged on postoperative day 7. The patient continued follow-up with regular dressings. After two months of follow-up, the patient remained asymptomatic. Informed consent was obtained from the patient and guardian for both the surgical procedure and the publication of this case report and related images.

DISCUSSION

Pilonidal sinus gets its name from the Latin words pilus (hair) and nidus (nest), reflecting the nature of the condition. Although the disorder was initially documented by Mayo OH in 1833, the term 'pilonidal disease' was later introduced by RM Hodges in 1880. During wartime, many US soldiers were diagnosed with pilonidal sinus, which was commonly referred to as 'Jeep disease' due to its high incidence among soldiers frequently travelling in jeeps [2].

Pilonidal sinus is an acquired condition, postulating that hair is drawn into the skin and subcutaneous tissue through mechanical forces, leading to a foreign body reaction and subsequent granuloma formation [3]. The condition has an estimated incidence of 26 cases per 100,000 individuals, with a male-to-female ratio of approximately 2.2:1 [4]. Pilonidal sinuses predominantly occur in the sacrococcygeal region of young adult males. Their association with the anal canal, however, represents a rare and atypical presentation. Fewer than 20 cases of perianal pilonidal sinus have been documented in the literature to date [5]. Aggarwal K et al., reported a case of pilonidal sinus originating in the intersphincteric region of the anal canal [6]. In contrast, the present case involves a pilonidal sinus located in the supralevator space, without any involvement of the anal sphincters.

Pilonidal sinus has been reported to affect a variety of other anatomical sites such as the breast, umbilicus, scalp, and penis [7]. Moreover, the aetiopathogenesis of this case was more akin to that of an anal fistula. Several cases in the literature have reported the development of complicated anal fistulas as a complication of anal abscesses, with the underlying aetiology being identified as anal pilonidal sinus during surgical intervention [5]. A thorough clinical evaluation is essential in patients presenting with a perianal fistula to distinguish between underlying conditions such as abscess formation, pilonidal sinus, and fistula-in-ano [8]. Accurate preoperative diagnosis is crucial, as misdiagnosis may result in inappropriate surgical planning, incomplete excision, or an increased risk of recurrence [9]. Differentiating between fistula-in-ano and pilonidal sinus can be accomplished through examination under anaesthesia or with the aid of MRI, the latter of which has proven to be a valuable tool in delineating complex perianal conditions and fistulous tracts [8].

Although pilonidal sinuses are typically confined to the subcutaneous tissue and perianal fistulas are anatomically connected to the anal canal or rectal mucosa, certain cases have demonstrated that infection originating from a pilonidal sinus can extend into the intersphincteric or even ischiorectal spaces, with fistulous openings occasionally reaching the natal cleft. Thus, MRI provides valuable information in such cases [10]. MRI is sensitive in differentiating between pilonidal sinus and fistula-in-ano, indicating that it may equally and accurately diagnose sepsis in both situations [11]. It has also been noted that while pilonidal sinus and perianal fistula have comparable MRI characteristics, pilonidal sinus is distinguished from the latter by the lack of an internal opening and intersphincteric sepsis [12]. A study has shown that preoperative MRI is considered the gold standard of care when diagnosing perianal fistulas [4].

However, in the present case, MRI failed to differentiate between fistula-in-ano and pilonidal sinus due to overlapping imaging characteristics. Both conditions can present as perianal sinus tracts with associated fluid collections and surrounding inflammation, particularly when the pilonidal sinus extends deeply or lies close to the anal canal. The tract in this case extended into the supralevator space, mimicking a supralevator fistula on imaging. Additionally, the presence of hair—considered a hallmark of pilonidal sinus—cannot be detected on MRI, further limiting its diagnostic specificity in such cases [8]. Moreover, the absence of a clearly visualised internal opening—typically present in fistula-in-ano—can be challenging to identify when local inflammation or oedema obscures the fine anatomical structures. This highlights the limitations of MRI in certain atypical or rare presentations and underscores the importance of intraoperative assessment and histopathological confirmation for accurate diagnosis.

Intraoperatively, the presence of hair shafts and keratinous debris within the fistulous tract was pivotal in prompting reconsideration of the diagnosis. These findings are pathognomonic for pilonidal sinus and differ significantly from the typical features of a cryptoglandular fistula-in-ano. Recognising these intraoperative hallmarks allowed for complete excision and histopathological confirmation, ultimately preventing mismanagement [13].

Complete excision of the tract with healing by secondary intention was selected in this case due to the localised nature of the lesion, the absence of sphincter involvement, and the need to obtain a definitive histopathological diagnosis. Secondary healing allows for better wound drainage and reduces the risk of recurrence, particularly in cases where the diagnosis is uncertain preoperatively and atypical features are present, such as supralevator extension without anal canal communication.

While other methods like primary closure, Limberg flap, and Karydakis flap have shown good results in sacrococcygeal pilonidal sinus disease, they are less commonly employed in perianal regions due to their proximity to the anal sphincters and the risk of contamination [14]. Healing by secondary intention may be preferable in such anatomically complex or contaminated areas, as it avoids flap complications and allows for close postoperative monitoring. Moreover, in a series of atypical pilonidal sinus cases involving the perianal or perineal regions, wide local excision with secondary healing resulted in good outcomes without recurrence [15].

Author (Year)	Location of sinus	Patient Age/Sex	Symptoms	Preoperative diagnosis	Imaging used	Intraoperative findings	Management	Outcome
Kumar P et al., (2013) [17]	Perianal	42/M	Discharge, pain	Fistula-in-ano	Not specified	Hair in sinus tract	Wide local excision	Healed without recurrence
Aggarwal K et al., (2015) [6]	Intersphincteric region (anal canal)	40/M	Pain, perianal swelling	Anal fistula	Not mentioned	Hair and debris in tract	Excision+primary closure	Recovered, no recurrence
Eberspacher C et al., (2017) [18]	Anterior+posterior anal fistula	37/M	Discharge	Complex anal fistula	MRI	Pilonidal sinus tract with hair, posterior fistula	Two-step surgery	Successful
Sert OZ (2020) [5]	Perianal	31/M	Discharge, pain	Fistula-in-ano	MRI	Pilonidal sinus with no internal opening	Excision+secondary healing	Healed well
Abdelatty MA et al., (2024) [12]	Sacrococcygeal (MRI focus)	-	-	Pilonidal sinus	MRI	-	Not specified	High interobserver agreement
Present case (2025)	Left perianal with supralevator extension	17/M	Discharge, pain on defaecation	Supralevator fistula	MRI	Hair, pus in tract; no internal opening	Excision+secondary healing	Asymptomatic at 2-month follow-up

[Table/Fig-5]: Comparison of previously reported cases of atypical pilonidal sinus presenting as perianal or fistulous lesions [5,6,12,17,18].

Given the supralevator extension in this case, alternative treatment options such as a staged approach with seton placement, minimally invasive techniques like Video-Assisted Anal Fistula Treatment (VAAFT), or Endoscopic Pilonidal Sinus Treatment (EPSiT) may be considered in similar cases. These approaches aim to minimise tissue disruption and preserve sphincter function, especially when preoperative imaging suggests high or complex tracts extending above the levator ani muscle [16]. Similar cases from the literature have been tabulated in [Table/Fig-5] [5,6,12,17,18].

CONCLUSION(S)

Perianal pilonidal sinus is a highly rare condition in clinical practice. The diagnosis of such cases is often confused with fistula-in-ano or perianal abscess. Although rare, the possibility of perianal pilonidal sinus must be considered when diagnosing and managing perianal diseases, especially in young adult male populations. In our case, reaching the diagnosis of perianal pilonidal sinus was a challenge in itself, as both preoperative clinical and radiodiagnostic findings did not support the diagnosis. Hence, it was quite difficult to conclude that it was a case of perianal pilonidal sinus, which was only identified intraoperatively, coming as a surprise, and was later confirmed by histopathological findings.

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