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Research Article

Pilonidal Sinus Mimicking Fistula in Ano in a Male Patient. A Case Report and Review of Literature

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Abstract

Introduction: Pilonidal Sinus typically presents as an abscess, purulent discharge, or pits around midgluteal cleft. In rare cases sinuses progress anteriorly mimicking Fistulas in Ano. Clinicians and especially surgeons should be aware of this situation, in order to avoid reoperations and patient's discomfort and frustration.

Materials and Methods: A 41 years old male, presented at the outpatient Surgical Clinic of our hospital on Oct 2018, due to perianal pus irritation persistent for a decade. On clinical examination, we observed an external perianal opening draining pus, without other findings. A pelvis MRI followed for further investigation. A perianal fistula was demonstrated, and the patient was submitted to fistulectomy and Seton positioning. Two months later a fistula recurrence was observed and a second MRI was followed. The anal fistula was related with a misdiagnosed pilonidal sinus. Our patient undergone a second procedure for fistulectomy and a pilonidal sinus removal. Seven months later the third operation was accomplished and both the sinus and the fistula were treated successfully with FiLaC™ technique (Fistula - Tract Laser Closure). Patient had a follow up of a period of one year and he was in an excellent condition without symptoms of recurrence.

Results and Discussion: In rare situations Pilonidal Sinuses appear as Fistulas in Ano. Few cases are described in the literature but it seems to be a more common situation than we believe. There are three main theories explaining this incident.

Conclusion: Surgeons should be aware for this entity in order to avoid unnecessary reoperations and patient's dissatisfaction. Appropriate preoperative imaging (MRI-Magnetic Resonance Imaging or EUS- Endorectal Ultrasound) and a two step surgery are the main keys for a successful management.

Keywords: Anal Fistula; Pilonidal Sinus; Recurrence; Anal Fistula Treatment

Abbreviations

MRI: Magnetic Resonance Imaging; EUS: Endorectal Ultrasound; DRE: Digital Rectal Examination; FiLaC™: Fistula-Tract Laser Closure; SiLaC™: Sinus Laser Assisted Closure; LIFT: Ligation of Intersphincteric Fistula Tract; VAAFT: Video Assisted Anal Fistula Treatment

Introduction

Pilonidal Sinus has an estimated prevalence of 0.7%, affecting mainly males usually among 16 and 25 years old [1]. It was first described in 1833 by Mayo but the term pilonidal sinus was given by P.M. Hodges in 1880 [4,9]. Typically, a pilonidal sinus presents as an abscess, pus drainage or pits in the midline of natal cleft. Rarely the sinuses can tract anteriorly mimicking Fistulas in Ano. Until today only a few dozens similar incidents have been described in literature. In these cases, it is essential to diagnose any association between the fistula and pilonidal sinus preoperatively in order to avoid recurrences, unnecessary reoperations and patient's dissatisfaction.

Materials and Methods

A 41 years old man, visited the outpatient Surgical Clinic of our hospital on Oct 2018, complaining for perianal pus drainage for the last 10 years. He reported no other health problems, no medications or any previous surgery. During clinical examination we spotted an external perianal opening at the 2 'o clock position, with the patient in lithotomy position, approximately 4 cm from anal verge, without an internal opening or other findings on digital rectal examination (DRE). Hence, we arranged a pelvis MRI for further investigation. MRI revealed a perianal fistula in close contact with the external anal sphincter at 5 o' clock. Despite the report didn't referred to a potential Pilonidal sinus, careful review of the images revealed an area of high signal in the posterior perianal area (Figure 1-3). After informed consent, we proceeded on rectal examination under general anaesthesia. On anoscopy we found a superficial track originating from the outer opening (2' o clock) and was heading backwards in the left side of the anus towards 6 'o clock. From there originated a direct transphincteric track leading to an internal opening at 5 'o clock. We decided to proceed to a wide opening of the superficial cavity (that was initially seen in the MRI images Figures 2 and 3). Also, the fistula was dealt with a partial fistulectomy for the part of the tract situated externally to the external sphincter and with the placement of a loose Seton through the direct track for the transphincteric part (2/0 Ehtibond excel, Ethicon, J and J medical devices).

Figure 1: MRI 06/11/2018. Anal fistula tract.

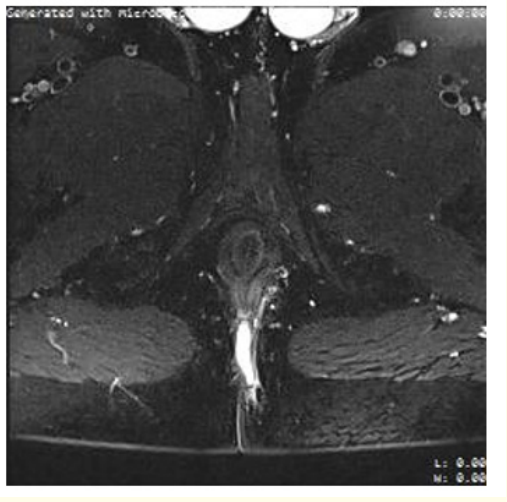

Figure 2: MRI 06/11/2018. Anal fistula tract.

Figure 3: MRI 06/11/2018. A potential pilonidal sinus.

Two months later, during the follow up, we spotted pus drainage from the point of the previous external opening. A second MRI of the area showed a fistula in ano communicating with the previous misdiagnosed pilonidal sinus (Figure 4). A second procedure was performed in order to handle the pilonidal sinus along with the recurrent discharge from the fistula. Before that, the patient was informed and a relevant consent for the combined procedure was taken. Also, preoperatively the anaesthetist and theatre stuff were

informed and an intraoperative change in position was planned from lithotomy to left lateral position. During the examination of the Rectum under Anaesthesia, in the posterior superficial perianal area a cavity was found extending between the external opening at 6 'o clock and the pilonidal sinus. Therefore, we decided to perform a pilonidal sinus excision with partial wound closure and a fistulotomy with marsupialisation of the superficial track connecting the cavity with the sinus. During this operation the previously placed loose Seton was left in place. Pathologic Examination of the midgluteal cleft specimen confirmed the diagnosis of the pilonidal sinus.

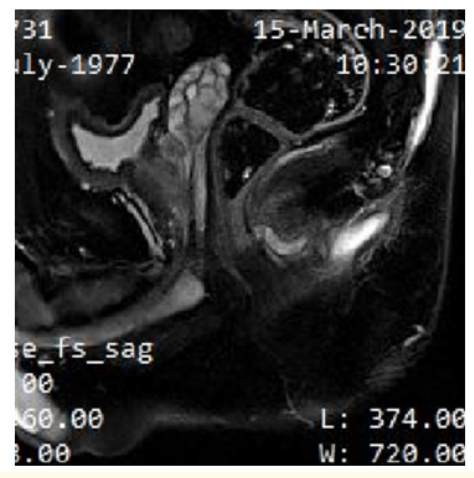

Figure 4: MRI 15/03/2019. MRI demonstrates a potential communication among the anal fistula and pilonidal sinus.

Our patient remained without symptoms for seven months but at that time, during of a schedule follow up check we noticed a recurrence of a small residual cavity in the sacrococcygeal area so a third operation was mandatory. During the procedure a probe was used to reveal the connection of the remaining pilonidal sinus cavity with the area of the external opening of the fistula. Initially the remaining cavity was excised to sacrococcygeal fascia and the small resulting wound was left open to heal by secondary intention. Then the tract connecting the fistula with the sinus and the fistula tract bearing the Seton was treated with FilaC™ technique, by using a laser emission probe to destroy the epithelium of the fistula track. Postoperatively the patient had an uneventful recovery with minimal discomfort from the open wound which healed within three weeks. One year later the patient is in perfect condition, completely asymptomatic, without evidence of signs of recurrence neither in imaging (MRI) nor in clinical examination (Figure 5).

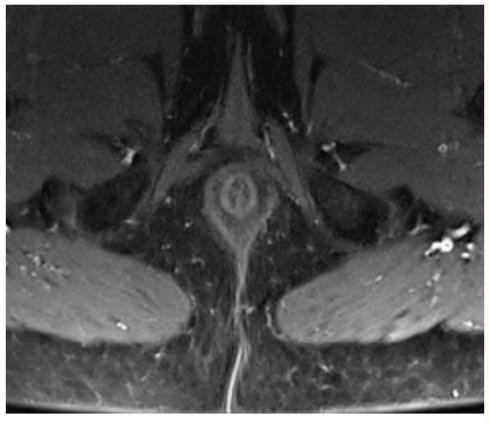

Figure 5: MRI 03/07/2020. Follow up after third surgery without imaging of remaining disease.

Results and Discussion

Pilonidal Sinus affects 21 per 100 000 people [2]. It's a common disease that affects mainly males between the ages 15 - 30 and less frequently after 60 years old [1,4,6]. Usually, it appears as an abscess or a drainage of midgluteal cleft [1,6]. Clinical examination regularly reveals pits on the sacrococcygeal region as a consequence of foreign body reaction - suppuration - and eventually track formation, or pus drainage in case of suppuration in acute phase. In rare occasions the tract doesn't forward to midgluteal cleft but to perianal region mimicking by this way fistulas in Ano. Few cases of simultaneously pilonidal sinus and fistula in Ano are described in literature but seems to be a more common situation than we believe [4,5]. Routine preoperative imaging of pilonidal disease is rarely necessary except in situations that we suspect an anorectal fistula, tumour or osteomyelitis where MRI and Ultrasound are mandatory [6].

Some factors that correlate with pilonidal sinus disease are hypertrichosis, deep gluteal cleft, overweight and seated position for hours. There are two main theories of sinus formation. The first one is that pilonidal sinus is a congenital disease caused by ectoderm fusion disorders during fetal development. The second one, which is the most accepted is the acquired theory. According to it, friction and microtraumatism of hair follicles in midgluteal cleft and traction of these follicles during gluteal movements create the conditions where growth of hair segments into subcutaneous tissue trigger an inflammation and foreign body reaction resulting to cyst formation. This theory strengthen from the fact that hairdress-

ers and animals' professionals (animal shearing) often suffer from fingers' hair follicles [6].

Pilonidal sinus treatment is mainly surgical. The aim of therapy is to remove the sinus and to have a rapid post operation recovery with less recurrences. The main therapeutic approaches are conservative techniques (simple incision and phenol treatment), excisional techniques without closure, excisional techniques with immediate closure (off midline closure, direct midline suture) and minimal invasive techniques (Sinus Laser assisted closure- SiLaC™; Biolitec, Video-assisted ablation) [14]. It's worth to notice that, in acute phase a simple incision is therapeutic in 58 - 82% of patients [6,13].

Fistulas in Ano appear in 8,6 people per 100,000, and it's prevalence is about 6.6 times higher in men [8,12]. About 90 - 95% of these fistulas originate from anorectal abscesses. According to the cryptoglandular theory, anorectal glands' inflammation and suppuration is the first step for anorectal abscess formation. Then the abscess progresses, creating a track which drains into the rectal or to perianal skin producing anal fistulas [2]. Other less common causing factors of anorectal fistulas are inflammatory bowel disease (Crohn's disease), prior operation, radiation, trauma, malignancy, infections (tuberculosis) and in seldom circumstances pilonidal sinus disease [2,3,8]. Clinical examination for Fistula in ano usually reveals one or more external openings and one or more internal openings. Preoperative imaging investigation (MRI or endorectall ultrasound) is useful in order to map the fistula track, and reveal associate fistulas or underlying diseases [11,12].

Anorectal fistulas' treatment constitutes a challenge for general surgeons due to high recurrence and complications rate. Many procedures have been described with different range of recurrences. Fistula's surgical cleaning and use of Seton is one of the most acceptable technique or the first step for other minimally invasion procedures. Fistulotomy can be therapeutic when is performed for low or simple fistulas. Nowadays several sphincter preserving techniques are developing with satisfactory results (heal rates from 69.7% to 82%) [15], like ligation of the intersphicteric fistula tract -LIFT, VAAFT (Video Assisted Anal Fistula Treatment), FiLaC™ (Fistula Tract Laser Closure), Anal fistula plugs, Radiofrequency and Rectal advancement flaps [12].

From the above it is clear that the diagnosis and treatment of pilonidal sinus disease and anal fistula are well described in the literature. But how do you deal with situations that pilonidal disease and anal fistula co-exist?

Few dozen cases of pilonidal disease mimicking fistulas in ano have been described. First of all is mandatory to make the proper diagnosis preoperatively. As we described above a pelvis MRI scan or endorectal ultrasound are the imaging of choice to investigate an anal fistula not only for having the accurate diagnosis but also to exclude a possible underlying disease. On clinical examination some findings will increase our suspicion that anal fistula is a result of pilonidal sinus. An obvious or hidden pilonidal disease, hair on the outer anal opening, the absence of primary fistula's opening and the absence of a palpable track connecting the external opening with the anus in the Digital Rectal Examination, could be indications that the anal fistula is associated with a Pilonidal sinus [10].

Currently there are three main theories in the literature that can explain this entity. The supporters of the first one believe that in these patients a pilonidal sinus and a fistula coexist, and their tracts accidentally meet each other. According to the second theory a perianal abscess pre-exists and a track is expanded posterior to the sacrococcygeal region constructing a cavity hence mimicking a Pilonidal Sinus. The advocators of the last theory, believe that a Pilonidal Sinus pre-exist and a track origins from the cavity progresses posteriorly to the perianal area mimicking fistula in ano [2,8]. In our case we believe that a pilonidal sinus and a fistula in ano coexisted as we could notice from the first imaging examination (Figure 2). We also consider that the misdiagnosed sinus at the first operation was the potential causing factor of the fistula's failure to mature despite the successful positioning the Seton. As a result the appropriate therapy was completed only after pilonidal sinus treatment.

According to the literature the most preferred treatment strategy in these patients is a two-step procedure. First step's aim is the drainage of abscesses, curettage of every cavity, excision of pilonidal sinus and Seton positioning in the fistula track. The second step is necessary for re-evaluation, removing the Seton and completion of the fistula treatment with the appropriate method. In our opinion the minimal invasive techniques like FilaC, or VAAFT are feasible, effective (high succeed rates) with low recurrence rate and morbidity and can be used successfully in one setting for the treatment of both the fistulas and sinuses in these cases [3,12].

Therefore, these situations are challenging for General Surgeons both for their diagnosis and-treatment. The most important first step is to have a high suspicion preoperatively with an accurate and detailed clinical examination, not only of the perianal but also of the sarcococcygeal area. In ambiguous cases an accurate preop-

erative imaging with MRI scan and Endo Anal Ultra Sound (EAUS) (whenever available), providing to the radiologist all the necessary clinical information can be valuable to the accurate preoperative diagnosis and correct planning of the treatment. It's important to notice here that our patient had a preoperative imaging that was not accurate about the presence of the pilonidal sinus and the potential communication with the perianal fistula, something that is not surprising as this information wasn't specifically requested by the radiologist who also lacked the relevant clinical information. As a result, our patient had to undergone a second operation in order to identify this association and complete the appropriate surgical treatment.

MRI scan and endorectal ultrasound (accuracy 80 - 90%) are the imaging examinations of choice to investigate a perianal fistula preoperatively [7,11]. It's crucial for every patient with preoperative suspicion of complex or secondary fistula before any procedure of fistula in ano, to have at least an imaging of rectum and perianal area in order to demonstrate the fistula track and its association with surroundings tissues. By this way the surgeon has the advantage to inform the patient and the theatre stuff correctly in order to avoid intraoperative surprises. As a result, patient can understand the complexity and pathology of his/her disease better and be able to give the relevant consent for both procedures. Simultaneously the surgeon can inform the anaesthetist and theatre stuff in time in order to plan for the appropriate positioning of the patient to the operating table.

Conclusion

Both fistulas in Ano and Pilonidal Sinus are common situations especially in young males. Rarely a Pilonidal Sinus can appear as a fistula in ano, a situation that can cause patient's discomfort and lead to unnecessary reoperations: Detailed preoperative clinical examination can lead to early clinical suspicion of this situation and allow for early planning of the appropriate preoperative imaging. Then the good collaboration with the radiologists, who need to know all the necessary clinical findings and questions in advance, is essential for the imaging to provide the information required. Finally, the patient with the help of his surgeon needs to understand the dual nature of his condition and agree to a combined plan of action in order to avoid reoperations and have the best outcomes.

Conflicts of Interest

The authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

Bibliography

1. Lim J and Shabbir J. "Pilonidal Sinus Disease - A Literature Review". *World Journal of Surgery and Surgical Research* 2 (2019): 1117.

2. Jeremy Sugrue., *et al.* "Pathogenesis and persistence of cryptoglandular anal fistula: a systematic review". *Techniques in Coloproctology* 21 (2017): 425-432.

3. Chiara Eberspacher., *et al.* "Pilonidal disease mimicking anterior anal fistula and associated with posterior anal fistula: a two-step surgery". *Case Report. G Chir* 38.6 (2017): 313-317.

4. Corey W Iqbal., *et al.* "Pilonidal Disease Mimicking Fistula-in-Ano in a 15-Year-Old Female". *Hindawi Publishing Corporation, Case Reports in Surgery* (2012).

5. S Vallance. "Pilonidal fistulas mimicking fistulas-in-ano". *British Journal of Surgery* 69.3 (1982): 161-162.

6. V De Parades., *et al.* "Pilonidal sinus disease". *Journal of Visceral Surgery* 150.4 (2013): 237-247.

7. Stuart A Taylor., *et al.* "Pilonidal Sinus Disease: MR Imaging Distinction from Fistula in Ano". *Radiology* 226 (2003): 662-667.

8. Pankaj Garg. "Anal fistula and pilonidal sinus disease coexisting simultaneously: An audit in a cohort of 1284 patients". *International Wound Journal* 16.5 (2019): 1199-1205.

9. Giuseppe Accarpio MD., *et al.* "Pilonidal Sinus with an Anal Canal Fistula-Report of a Case". *Diseases of the Colon and Rectum* 31.12 (1988): 965-967.

10. Gerald Mason Feigen MD., *et al.* "Pilonidal Disease Simulating Rectal Abscess and Fistula". *The Art and Science of Surgery* 73.2 (1956): 258-260.

11. A Amato., *et al.* "Evaluation and management of perianal abscess and anal fistula:a consensus statement developed by the Italian Society of Colorectal Surgery (SICCR)". *Techniques in Coloproctology* 19 (2015): 595-606.

12. Matthew Sammu Paul Skaife. "The management of cryptoglandular fistula-in-ano". *British Journal of Hospital Medicine* 81.1 (2020).

13. Iesalnieks I and Ommer A. "The management of pilonidal sinus". *Deutsches Ärzteblatt International* 116 (2019): 12-21.

14. J Harju., *et al.* "Pilonidal Disease Treatment By Radial Laser Surgery (FilaC™): The First Finnish Experience". *Scandinavian Journal of Surgery* (2020): 1457496920975610.

15. H Elfeki., *et al.* "A systematic review and meta-analysis of the safety and efficacy of fistula laser closure". *Techniques in Coloproctology* (2020).

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