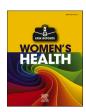
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Vaginocutaneous fistula caused by recurrent labial abscess: A case report

Komkrit Aimjirakul, Teerapan Seehanantawong, Apisith Saraluck

Division of Female Pelvic Medicine & Reconstructive Surgery, Department of Obstetrics & Gynaecology, Faculty of Medicine Ramathibodi Hospital, Mahidol University, 270 Rama VI Road, Rajataewe, Bangkok 10400, Thailand

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ABSTRACT

Urogenital fistulas are characterized by communication tracts that connect two surfaces or distinct organ systems. A vaginocutaneous fistula is a rare type of urogenital fistula that is characterized by persistent perineal discharge. This case report describes a female patient who suffered from recurrent labial abscesses and a fistula that connected the lower vagina to the inner thigh. There were symptoms of persistent perineal discharge, and the patient had an offensive odor. The patient had a history of six recurrent right labial abscesses over a period of two years prior to the formation of two fistulous openings. The right medial aspect of the inner thigh, adjacent to the labia majora, was found to have a 0.7 cm opening, which was confirmed by magnetic resonance imaging. Intraoperatively, the fistulous tract was readily delineated using dye. Complete fistulectomy was performed. This report highlights an unusual presentation of a urogenital fistula, the diagnostic challenges, and the management strategies. It emphasizes the need for timely identification and intervention in such cases.

1. Introduction

Fistulas are communication tracts that connect two surfaces or distinct organ systems. Fistulas are located internally or migrate outward, establishing a communication tract between the internal viscera and the skin surface. [1] Vaginocutaneous fistula describes a lesion that communicates from the vagina or vaginal canal to the epidermis, primarily in the labia major, vulva, groin or adjacent area. [2] These fistulas are extremely rare, with the majority of cases resulting from obstetrical injury. [3] This report concerns the case of a 44-year-old woman who developed a rare vaginocutaneous fistula as a result of multiple recurrent episodes of labial abscess. This report also presents a review of the limited literature on this unique entity.

2. Case Presentation

A 44-year-old nulliparous woman was referred to the urogynaecological clinic from the general gynaecological clinic for assessment of recurrent discharge from her right inner thigh, which had been present for 2 months. The volume (beginning with scant flow) and foul odor of the discharge had increased over time. The discharge was intermittently bloody and had a yellowish colour. Additionally, the patient experienced intermittent episodes of low-grade fever and localised discomfort.

The patient was otherwise healthy, with no underlying conditions,

except for six recurrent right labial abscesses over the course of 2 years. Antibiotics had been administered at each episode, followed by aspiration or incision and drainage, which led to symptomatic improvement. However, the abscess had recurred every 3–4 months.

The perineum was examined, and a 0.7 cm opening was found at the right medial aspect of the inner thigh, adjacent to the labia majora (Fig. 1). Additionally, a 0.2 cm opening was observed at the right site of the inner vaginal wall. The vestibule was otherwise unremarkable, and there were no other obvious external openings. No signs of infection or inflammation were apparent in other areas of the patient's body, and no lymph nodes were enlarged. The opening inside the patient's vagina bled a small amount as a result of manipulation of the surrounding tissue. However, there was only a small amount of yellowish pus. The remaining parts of the clinical examination were unremarkable.

Magnetic resonance imaging revealed a fistulous tract between the right side of the lower vagina and the posteroinferior aspect of the inner thigh next to the labia majora. The length of the tract in the coronal plane or craniocaudal direction was approximately 3.7 cm. Marked inflammation was visible around the tract (Fig. 2). A clear tract to the anal canal, or a buttock mass or pelvic lymph node enlargement was not evident. The results of the preoperative investigations were within normal limits.

The fistulous tract was readily delineated intraoperatively using dye and probe insertion. There was a single tract without branching or occult

E-mail addresses: abhisith.13@gmail.com, apisith.sa@mahidol.ac.th (A. Saraluck).

^{*} Corresponding author.



Fig. 1. Vaginocutaneous fistula connected between the right medial aspect of the inner thigh.

communications. An incision was created around the fistulous tract, using the probe as a guide. The fistula opening was completely excised, with healthy margins, followed by excision of the fistulous tract itself (Fig. 3). The subcutaneous tissues and skin were then closed.

The patient was discharged after the urethral catheter and vaginal tampon were removed on the first postoperative day. A wound swab obtained earlier showed growth of mixed flora with contaminants; hence, broad-spectrum antibiotics were continued, and the patient was advised to perform daily sitz baths and ensure perineal hygiene. The water may be plain or contain substances like salt, baking soda, or

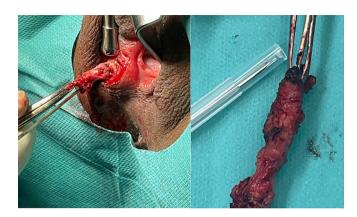


Fig. 3. Intra-operative image of the fistulectomy.

antiseptic solutions, depending on the intended therapeutic effect.

The postoperative period was uneventful, and at 1, 3 and 6 months of follow-up, the wound showed complete healing. The histological report indicated acute and chronic inflammation with granulation tissue from the sinus tract.

3. Discussion

Vaginocutaneous fistula is the most uncommon type of genital fistula, with fewer than 10 cases documented in the literature. [3,4] Additionally, vaginocutaneous fistulas have been reported to develop as complications of postpartum-related conditions or urogynaecological surgery, including sacrospinous fixation or anti-incontinence procedures. [5] Patients typically experience dyspareunia, pain and

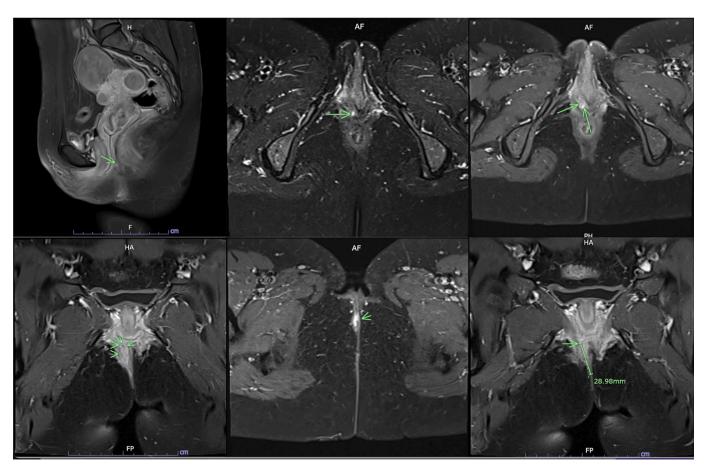


Fig. 2. Magnetic resonance imaging (MRI) revealed a fistulous tract.

unpleasant-smelling discharge from the vagina or surrounding skin, as well as systemic symptoms such as fever and fatigue, which are secondary to infection. [1]

Factors that influence wound healing and vascularity are the primary risk factors associated with genital fistulas, including vaginocutaneous fistulas. Poor nutritional status, comorbidities (such as diabetes mellitus and immunosuppression) and postmenopausal status with oestrogen deficiency are among the primary general factors that compromise tissue integrity. [6] Vascularity-related concerns include connective tissue diseases, malignancies, localised infections, pelvic irradiation and previous pelvic surgeries. Reactions to a foreign body, erosions caused by synthetic materials and tension produced by rigid sling insertion are all factors that can be observed during mesh-related surgical procedures. [7] Additionally, obstetrical factors include traumatic childbirth and macrosomia, protracted second stage of labour and trauma from an obstetrical procedure or episiotomy incision. [8] Healing is considerably impeded by each of these factors, which increases the likelihood of fistula formation. Interestingly, in this case, the patient had no history of pregnancy or vaginal delivery and no history of urogynaecological procedures. The only possible risk factor in this patient was previous multiple episodes of right labial abscess with a history of incision and drainage. Even though incision and drainage is not an invasive urogynaecological procedure, possible factors induced by the infection, inflammation leading to granulation and the healing process likely led to the development of the fistulous tract in this patient.

Previous reports from India have documented the development of a vaginal fistula following a normal vaginal delivery with episiotomy, with an onset of 2–3 weeks. [5,9] Perineal tears resulting from traumatic delivery and non-healing of the episiotomy wound occasionally lead to fistulas. The reports from India addressed not only surgical management but also infection management, including delayed suturing of the wound and the prescription of broad-spectrum antibiotics. [5,9]

Almost all previous reports have documented the development of vaginocutaneous fistulas following urogynaecological procedures. In some reports, the fistula developed after transobturator tape and tension-free vaginal tape surgeries for stress urinary incontinence. [10,11] The inflammatory response induced by propylene monofilament sling mesh may result in erosion of adjacent tissue, which is another risk factor for fistula formation. Notably, one report of two patients with an immunological condition (Sjögren's syndrome) has been published. [12] Both cases were treated surgically, which involved the removal of the mesh, debridement of fistulous tissue and abscess drainage. Moreover, a case of a cutaneous gluteal-vaginal fistula was reported following sacrospinous fixation, in which two nonabsorbable sutures were inserted into the right sacrospinous ligament. The patient was successfully managed through computed tomography-guided percutaneous drainage of the gluteal abscess and placement of a guiding cutaneous vaginal catheter, laparoscopic pelvic wall dissection and evaluation. [13]

Inflammatory bowel diseases (IBD) like Crohn's can cause vaginocutaneous fistulas because of the constant granulomatous inflammation and involvement of transmural tissue. Biologic therapy, such as anti-TNF inhibitors, may be necessary for IBD-related fistulas, in contrast to those caused by surgical or infectious complications. In some suspected cases, accurate diagnosis requires histological findings, imaging investigations, and gastrointestinal evaluation. [14]

The investigation of vaginocutaneous fistulas involves magnetic resonance imaging to localise and delineate the tract, as well as any branches or communications, with an emphasis on the exclusion of an anorectal fistula. [15] Fistulography, dye installation with methylene blue and probe insertion are the most cost-effective diagnostic approaches, although are less sensitive than other methods. The dye is introduced through the external opening and the internal opening(s) is identified by dye overflow. [9]

The management of vaginocutaneous fistulas involves complete excision of the tract by coring from the outside inward. To facilitate

wound healing, it is essential to prescribe appropriate antibiotics and to apply dressings. One of the post-operative care strategies is a warm sitz bath. [16] A warm sitz bath is a shallow bath in which a person sits in warm water, typically covering only the hips and buttocks. It is often used to cleanse the perineal area, promote healing, and provide relief from discomfort caused by conditions such as perineal wounds, hemorrhoids, or post-surgical recovery. [17] Alternative treatments are similar to those used for an anal fistula, including laser or coagulation. [1] There is also discussion regarding novel alternative treatment options, including platelet-rich plasma injection, radiofrequency ablation and cyanoacrylate adhesive glue. However, these are not considered standard and require additional research. [18–20]

4. Conclusion

In conclusion, in this case a vaginocutaneous fistula which had developed as a result of multiple recurrent labial abscesses was successfully identified and managed. In patients with potential risk factors, clinical suspicion of rare consequences, such as a cutaneous fistula, should be considered in the differential diagnosis, even if the situation is as simple as a recurrent abscess and post-incision and drainage. A holistic approach and a multidisciplinary team should be implemented to enhance the patient's quality of life and achieve a positive outcome.

Contributors

Komkrit Aimjirakul contributed to patient care, acquiring and interpreting the data and revising the article critically for important intellectual content.

Teerapan Seehanantawong contributed to patient care, acquiring and interpreting the data and revising the article critically for important intellectual content.

Apisith Saraluck contributed to patient care and the conception of the case report, drafting the manuscript, undertaking the literature review, interpreting the data and revising the article critically for important intellectual content.

All authors approved the final submitted manuscript.

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Patient consent

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Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

References

- [1] S. Carr, A.L. Velasco, Fistula-in-Ano, in: StatPearls, StatPearls Publishing Copyright
 2024, StatPearls Publishing LLC, Treasure Island (FL), 2024.
- [2] O. Chinthakanan, P. Sirisreetreerux, A. Saraluck, Vesicovaginal fistulas: prevalence, impact, and management challenges, Medicina 59 (11) (2023) 1947.
- [3] R.G. Rogers, P.C. Jeppson, Current diagnosis and Management of Pelvic Fistulae in women, Obstet. Gynecol. 128 (3) (2016) 635–650.
- [4] H.W. Brown, L. Wang, C.H. Bunker, J.L. Lowder, Lower reproductive tract fistula repairs in inpatient US women, 1979-2006, Int. Urogynecol. J. 23 (4) (2012) 403-410
- [5] A. Ramji, Vagino-cutaneous fistula: unreported, under-reported or unheralded? Int. J. Reprod. Contracept. Obstet. Gynecol. 8 (2019) 2932.
- [6] G. Gethin, Understanding the inflammatory process in wound healing, Br. J. Community Nurs. (2012) Suppl:S17-8, s20, s2. volumn17 No.sub3 Page 1-3.
- [7] N.X. Landén, D. Li, M. Ståhle, Transition from inflammation to proliferation: a critical step during wound healing, Cell. Mol. Life Sci. 73 (20) (2016) 3861–3885.
- [8] G. Dorairajan, N H., Chronic non-healing sinus manifesting in episiotomy scar: hidden fistula-in-ano, Int. Urogynecol. J. 25 (10) (2014) 1441–1443.
- [9] Y. Ohara, T. Enomoto, Y. Owada, K. Hisakura, Y. Akashi, K. Ogawa, et al., Rectoperineal fistula presented 5 months after repair of severe obstetric perineal laceration: a case report, Front. Surg. 8 (2021) 637719.
- [10] A.F. Şahin, Y. İlbey, N. Şahin, Vaginocutaneous fistula and inguinal abcess presented 6 years after tension-free vaginal tape sling, Arch. Ital. Urol. Androl. 85 (2) (2013) 104–106.
- [11] M. Maffiolini, C.R. Asteria, A cutaneous-vaginal fistula and myositis of the obturator muscle following placement of a trans-obturator tape for stress incontinence, Eur. J. Obstet. Gynecol. Reprod. Biol. 149 (2) (2010) 225–226.

- [12] K. Beksac, M. Turgal, D. Basaran, O. Aran, M.S. Beksac, Vaginoperineal fistula as a complication of perianal surgery in a patient with Sjögren's syndrome: a case report, Case Rep. Rheumatol. 2014 (1) (2014) 359605.
- [13] V. Kim, S. Seraji, B.A. Grigorescu, M. Hon, D.H. Hunt, F.R. Nezhat, Multidisciplinary Management of Cutaneous Gluteus Vaginal Fistula after Sacrospinous Ligament Fixation, Crsls 10 (1) (2023).
- [14] J. Panes, W. Reinisch, E. Rupniewska, S. Khan, J. Forns, J.M. Khalid, et al., Burden and outcomes for complex perianal fistulas in Crohn's disease: systematic review, World J. Gastroenterol. 24 (42) (2018) 4821–4834.
- [15] D.L. Giles, G.W. Davila, Suprapubic-vaginocutaneous fistula 18 years after a bladder-neck suspension, Obstet. Gynecol. 105 (5 Pt 2) (2005) 1193–1195.
- [16] W.C. Liao, Y.Y. Cheng, C.K. Hsu, Y.C. Chiu, H.Y. Chiu, S.C. Chang, et al., Effects of early warm water sitz bath on urinary retention and pain after haemorrhoidectomy: a randomized controlled trial, Int. J. Nurs. Stud. 154 (2024) 104765.
- [17] A. Shafik, Role of warm-water bath in anorectal conditions. The "thermosphincteric reflex", J. Clin. Gastroenterol. 16 (4) (1993) 304–308.
- [18] P.J. Gupta, Radio frequency "sutureless" fistulotomy- a new way of treating fistula in anus, World J. Gastroenterol. 9 (5) (2003) 1082–1085.
- [19] G. Romero, C. Santos, P. Cassino, M. Odashiro, G. Seno, G. Aléssio, Treatment of fistula-in-ano with cyanoacrylate glue with and without prior seton placement in rats, Acta Cir. Bras. 31 (2016) 377–381.
- [20] A. Saraluck, O. Chinthakanan, A. Kijmanawat, K. Aimjirakul, R. Wattanayingcharoenchai, J. Manonai, Autologous platelet rich plasma (A-PRP) combined with pelvic floor muscle training for the treatment of female stress urinary incontinence (SUI): a randomized control clinical trial, Neurourol.Urodyn. 43 (2) (2024) 342–353.