Electronic Journal of Medical and Dental Studies
2023, 13(1), em0103
e-ISSN: 2753-2658
https://www.ejmds.com

MODESTUM

Letter to the Editor

OPEN ACCESS

Perineal scar endometriosis

Carl Abi Semaan [1], Amarelle Chamoun [2], Jad Ghauch [1], Hiba Hamdar [3]*

[1] University Saint Esprit Kaslik, Jouniyeh, LEBANON
[2] Faculty of Medical Sciences, Lebanese University, Beirut, LEBANON
[3] Plovdiv Medical University, Plovdiv, BULGARIA
***Corresponding Author:** hiba.hamdar@live.com

Citation: Abi Semaan C, Chamoun A, Ghauch J, Hamdar H. Perineal scar endometriosis. ELECTR J MED DENT STUD. 2023;13(1):em0103. https://doi.org/10.29333/ejmds/12813

ARTICLE INFO

Received: 15 Dec. 2022

Accepted: 03 Jan. 2023

ABSTRACT

The presence of ectopic endometrial tissue outside the uterine cavity is referred to as endometriosis. It is a harmless condition that is commonly seen in women of reproductive age. It can happen in both the pelvic and extra-pelvic areas. Endometriosis in an episiotomy scar is extremely rare, but due to local infiltration, it can cause significant morbidity in patients. The presence of the classic clinical triad of episiotomy history, tender nodule at the scar site, and cyclical pain can be used to diagnose this condition. Magnetic resonance imaging is a very useful imaging modality for diagnosing and assessing the lesion's deeper extent.

Keywords: perineal scar endometriosis, aetiology, pathogenesis, lesions, diagnosis, surgery

INTRODUCTION

Endometriosis is a benign condition that typically affects the pelvic area and is linked to an inflammatory process that is oestrogen-dependent and causes the formation of endometrial tissues and glands outside the uterine cavity. Endometriosis can affect the umbilicus, abdominal wall, lungs, or even the vulva or perineum outside of the pelvic area [1]. Its aetiology is unknown, but it involves complex interactions between genetic makeup, hormonal functionality, menstrual cycle, different levels of inflammation, and immunological components [2]. It affects about 10% of reproductive-age women and 35-50% of women with infertility and chronic pelvic pain. While extra-perineal endometriosis (PEM) is less common than uterine endometriosis, accounting for only 12% of cases, perineal scar endometriosis is even less common, accounting for only 0.03% to 0.15% of all cases [3, 4].

The pathogenesis of endometriosis has been the subject of numerous theories, some of which include retrograde menstruation or the implantation of uterine endometrial cells via lymphatic dissemination, direct implantation, haematogenous spread, or coelomic metaplasia. The mechanical transplantation of endometrial cells in the months following an open episiotomy scar induced by vaginal delivery is thought to be the cause of episiotomy scar endometriosis [5]. A history of episiotomy, tender nodules at the scar site, and menstrual pain are the typical clinical triad of symptoms of perineal scar endometriosis. These symptoms are only observed in half of the cases, resulting in frequent misdiagnosis. As a result, MRI is required to make a final diagnosis and characterize the severity of the condition. The primary treatment for perineal scar endometriosis is surgical excision with clear wide margins [3].

DISCUSSION

PEM, also known as episiotomy scar endometriosis, was first described in 1923 and is defined by the presence of endometrial tissue in the perineal region, in either the vulva or subcutaneous tissue. Anal sphincter involvement is seen in approximately half of the patients, resulting in dyspareunia, faecal incontinence, and surgical fistula. As a result, early detection and treatment are critical [1, 6, 7]. PEM is an extremely rare disease that affects only 0.3% to 1% of women [8]. Vulvo-PEM is classified into two types: cystic and nodular lesions, each with its own aetiology and treatment options [4]. PEM is most common after a vaginal delivery. It can also occur following Bartholin gland excision sites within the vulva [9]. Other factors that contribute include lymphatic dissemination, haematogenous spread, familial factors, immunological factors, and mullerian remnant metaplasia. As a result, the proclivity for PEM is polygenic and multifactorial. According to one study, having a first-degree relative with endometriosis increases a woman's risk of developing the condition seven times [10].

Despite the fact that perineal trauma is the most common cause of vulvo-PEM, scar endometriosis can also be caused by other general surgical procedures such as appendectomy, inguinal hernial repair, laparoscopic cholecystectomy, or laparoscopic gastric by-pass. However, the occurrence of PEM following these surgeries is extremely rare [1]. Undifferentiated stem cells may be the underlying cause of distant endometriosis lesions [2]. The body mass index BMI is one of several risk factors for the development of PEM and can affect its incubation period.

According to research, the higher the BMI during delivery and within one month afterward, the shorter the incubation period of PEM. This is explained by the increased presence of fat in the perineum and lochia in patients with a high BMI, which leads to blood accumulation in the vagina and perineum, and thus endometrial cell implantation in the perineal incision. Furthermore, steroid sulfatase and 17 beta-hydroxysteroid dehydrogenases in ectopic endometrial cells activate sulphated steroid, the inactive oestrogen secreted by adipocytes [2, 11]. As a result, oestrogen's local action is amplified, resulting in a positive feedback loop that stimulates the growth of endometrial cells in the perineum. The age of onset, delivery age, breastfeeding period, and time of return to menstruation, on the other hand, have no effect on the incubation period of PEM. The duration of the pain, the size and number of nodules, involvement of the perianal muscles, and pelvic pain were all found to be unrelated to the pain in the lesions [2]. Other factors, such as genetic predisposition, immunological and familial factors, may also contribute to PEM pathogenesis [2, 10]. PEM is classified as primary or secondary, and its aetiology is unknown. Many theories, however, have been put forth, including direct implantation, which describes secondary PEM, retrograde menstruation, lymphatic dissemination, coelomic metaplasia, or haematogenous spread, which are signs of primary PEM [2, 5]. Endometrium may implant into the perineum episiotomy scar during vaginal delivery and grow into a PEM lesion, according to the "implanted theory" [12]. Endometrial cells may disappear spontaneously during vaginal birth, but they can also develop into endometriomas through cyclic recurrence, which causes clinically unpleasant symptoms [13]. It is important to note that simply transplanting these cells is insufficient because, as with peritoneal endometriosis, only a minority of pregnant patients with episiotomy or caesarean section develop endometriosis, implying that there are still unknown individual factors that favour cell transplant acceptance [1]. PEM, on the other hand, can be found on the opposite side of the episiotomy scar, and even nulliparous women can be diagnosed with it.

Another theory is retrograde menstruation, in which menstrual endometrium fragments pass through the fallopian tubes and implant and persist on peritoneal surfaces, which could explain why the Douglas cul-de-sac, ovaries, and uterus continue to be the primary sites of endometriotic lesions. However, neither of these theories can account for the disease's various locations [6]. Furthermore, the transplantation hypothesis explains how perineal trauma, such as perineal tearing or episiotomy after vaginal delivery, can result in PEM [11]. However, cases of PEM without anterior vulvo-vaginal trauma have been reported, in which retrograde menstruation metastases through the peritoneum with lymphovascular dissemination to the lungs, GI tract, perineum, and vagina, a phenomenon known as the "metastatic theory" [14]. This theory explains the pathogenesis of endometriosis lesions that develop spontaneously. This theory explains the pathogenesis of endometriosis lesions that develop spontaneously. Endometriosis in the labia majora may be caused by the spread of pelvic endometriosis, but it can only be attributed to coelomic metaplasia in Bartholin's gland [2]. Another proposed theory is the transformation of pluripotent peritoneal mesothelium. Furthermore, a neurologic theory recently proposed that the lesions migrate from the site of the original lesions and invade the large bowel along the nerves [10].

Perineal scar endometriosis has specific diagnostic criteria that, if found in a patient, guarantee a 100% accuracy in diagnosing the condition. The following are the criteria: a history of vaginal delivery perineal episiotomy, a painful mass or nodule at the perineum, and cyclic perineal pain during menstruation [15]. Early diagnosis is critical because delayed diagnosis can result in malaise caused by movements of surrounding structures during defecation, such as the anal sphincter and the rectum [3]. Patients frequently present to their healthcare providers after experiencing several months of perineal pain and swelling. The agonizing condition significantly disrupts the patients' daily lives; several reported continuous discomfort throughout the days that was not relieved by pain medication, as well as dyspareunia in some cases [16]. A large proportion of those affected have had an episiotomy following a vaginal delivery in the previous months or even years, whereas nulliparous women suffer from perineal scar endometriosis at a much lower rate.

Perineal scar endometriosis should be considered in a nulliparous patient who has nodules at the perineum that swell and hurt during menstruation [17]. Perineal scar endometriosis should be considered in a nulliparous patient who has nodules at the perineum that swell and hurt during menstruation. Despite the fact that endometriosis is an oestrogen-dependent inflammation that occurs before or during menstruation, patients do not report a change in cycle length or flow volume. Endometrial stroma and glandular infiltrations were found at the superficial level of the muscles adjacent to the scar, including the levator ani and the external anal sphincter, on pathologic examination of the excised nodules. Some patients had slightly elevated levels of serum cancer antigen 125 (CA125) [2, 6]. Scar endometriosis at the perineum is frequently misdiagnosed because its symptoms are frequently confused with hypertrophic scar tissue, abscesses, granulomas, malignancies, metastatic carcinomas, hernias, desmoids tumours, hematomas, neuromas, and other more common disorders [18].

Physical examination and laparoscopy alone cannot determine the extent of perineal scar endometriosis within deep pelvic tissues. Transvaginal ultrasounds and rectal sonographies, for example, do not detect deep pelvic lesions within the affected structures. As a result, magnetic resonance imaging (MRI) is the most rigorous modality for this evaluation [9]. The masses appear multi-lobular on MRI, with inner haemorrhaging, which is typical of vulvar endometriosis. Invasion of surrounding muscles of the external anal sphincter, as well as more distal structures damaged by the disease, can also be clearly identified [8]. If not treated promptly, perineal scar endometriosis can spread to neighbouring structures, most notably the rectum and the perineal muscles: the levator ani and the external anal sphincter. Faecal incontinence may result from deterioration of these structures [6, 8].

PEM is usually diagnosed based solely on a history and a physical exam. The sporadic growth of the lesion and discomfort during menstruation are diagnostic indicators. When patients first present, the majority have a sensitive, palpable subcutaneous swelling next to or inside the surgical scar [11, 13]. Because the typical symptoms are only present in 50% of patients, the presence of the traditional symptoms allows for the clinical diagnosis of perineal scar endometriosis [3, 6], and imaging is helpful in the diagnosis. In addition, imaging aids in determining the extent of surgical treatment in cases of larger lesions and in the preoperative assessment of

sphincter involvement, as well as ruling out other possibilities such as keloid, hematoma, granuloma, abscess, cysts, and tumour [5, 10, 19], but also anal fistula, atheroma, and hidradenitis [6, 12], which are differential diagnoses for perineal scar [2]. Ultrasound is frequently used to diagnose perineal scar endometriosis as the first imaging technique [3, 10]. A hypoechoic or anechoic lesion with fine internal echoes may exist at the site of the scar [3, 8, 11]. Peripheral vascularity may be visible as well.

Transrectal ultrasound with a high-frequency probe can diagnose rectal, recto-vaginal, or recto-sigmoid endometriosis, but penetration is poor [3, 9]. However, computed tomography [3, 20, 21] and transvaginal ultrasound are less useful in assessing perineal scar endometriosis. Some case studies show that the high-frequency power doppler angiographic appearance can also be used to identify scar endometriosis. Furthermore, because serum CA125 levels are usually normal or slightly elevated, it does not appear to be a reliable predictor of PEM [11, 13]. MRI is a great tool for determining the extent of a localized disease before surgery because it is a non-invasive imaging technique [8, 10]. It also has a high spatial resolution, which allows for good tissue characterization and multiplanar evaluation, and because it is highly sensitive, it can distinguish endometriomas from nearby tissue [22]. Nonetheless, it was highlighted utility of MRI in the preoperative evaluation of this condition in their report on a case of perineal scar endometriosis with anal sphincter involvement [3]. Endometriosis is also more likely to be detected when T1 and T2 hyperintensities are present, but saturation is absent [8]. Due to its excellent contrast resolution, MRI can determine size of the lesion and its relationship to the anal sphincter complex.

Endometriosis in the perineal scar is best treated with surgical excision with wide, clear margins. Recurrence is a possibility. As a result, complete removal of the lesion is critical. According to [3], the only way to prevent recurrence is to completely remove the lesion, including healthy margins [1, 2, 5, 9, 10, 22, 23]. Furthermore, any cystic lesions must be completely removed in order to prevent cyst fluid from infecting and implanting normal tissue [2]. Delaying surgery may cause the lesion to worsen and involve the anal sphincter [5, 24]. As a result, patients with anal sphincter involvement who did not receive complete excision experienced recurrence [23]. Following surgery, leuprolide, a gonadotropin-releasing analog, can be used to reduce the risk of recurrence [8, 24]. Hence, PEM patients treated with GnRH-agonists had a lower recurrence rate after surgery than PEM patients who did not receive GnRH-agonists. However, the decision to have PEM surgery should be made after considering the patient's expectations for the surgery's outcome, age, and desire for pregnancy. Furthermore, perineal scar endometriosis can be avoided by ensuring that the episiotomy scar is not contaminated with debris and blood. Gloves should be replaced before repairing an episiotomy wound [3]. Sphincteroplasty may be required to reduce the risk of faecal incontinence when the anal sphincter is affected.

CONCLUSION

Perineal scar endometriosis is a rare and harmless disease. To identify this illness in patients who present with no typical clinical symptoms, a high level of suspicion is required, and the earlier detection and intervention, the lower the morbidity and complications. Preoperative MRI, as well as ultrasound in some aspects of endometriosis, is extremely helpful in making the diagnosis and determining its local scope. Furthermore, they can rule out endometriosis in other areas of the pelvis. Following surgery, follow-up care and certain drug treatments are critical for preventing recurrence.

Author contributions: All authors have sufficiently contributed to the study and agreed with the results and conclusions.

Funding: No funding source is reported for this study.

Ethical statement: Authors stated that the consent was not required for this letter to the editor, which aims to spread the education regarding the subject between medical students and medical professionals as it is considered rare among the well known diseases and illnesses.

Declaration of interest: No conflict of interest is declared by authors.

Data sharing statement: Data supporting the findings and conclusions are available upon request from the corresponding author.

REFERENCES

1. Botezatu R, Turcu-Duminica A, Ciobanu AM, Gica N, Peltecu G, Panaitescu AM. Episiotomy scar endometriosis. Case presentation. Maedica (Bucur). 2021;16(4):713-6. https://doi.org/10.26574/maedica.2021.16.4.713
2. Liang Y, Zhang D, Jiang L, Liu Y, Zhang J. Clinical characteristics of perineal endometriosis: A case series. World J Clin Cases. 2021;9(5):1037-47. https://doi.org/10.12998/wjcc.v9.i5.1037 PMid:33644167 PMCid:PMC7896645
3. Jayanthan SS, Shashikala G, Arathi N. Perineal scar endometriosis. Indian J Radiol Imaging. 2019;29(4):457-61. https://doi.org/10.4103/ijri.IJRI_366_19 PMid:31949353 PMCid:PMC6958888
4. Maillard C, Cherif Alami Z, Squifflet JL, et al. Diagnosis and treatment of vulvo-perineal endometriosis: A systematic review. Front Surg. 2021;8:637180. https://doi.org/10.3389/fsurg.2021.637180 PMid:34046423 PMCid:PMC8148344
5. Dadhwal V, Sharma A, Khoiwal K, Nakra T. Episiotomy scar endometriosis. Med J Armed Forces India. 2018;74(3):297-9. https://doi.org/10.1016/j.mjafi.2017.06.004 PMid:30093779 PMCid:PMC6081211
6. Liu Y, Pi R, Luo H, Wang W, Zhao X, Qi X. Characteristics and long-term outcomes of perineal endometriosis: A retrospective study. Medicine (Baltimore). 2020;99(23):e20638. https://doi.org/10.1097/MD.0000000000020638 PMid:32502046 PMCid:PMC7306333
7. Eray IC, Topal U. Perineal scar endometriosis involving the anal sphincter. A case report and review of the literature. Ann Ital Chir. 2021;10:S2239253X2103173X.
8. Hakim H, Ben Halima S, Zouari A, et al. Perineal endometriosis: A rare case of a unique sizeable nodule. Pan Afr Med J. 2021;38:47. https://doi.org/10.11604/pamj.2021.38.47.27737 PMid:33854676 PMCid:PMC8017358
9. Matalliotakis M, Matalliotaki C, Zervou MI, Krithinakis K, Goulielmos GN, Kalogiannidis I. Abdominal and perineal scar endometriosis: Retrospective study on 40 cases. Eur J Obstet Gynecol Reprod Biol. 2020;252:225-7. https://doi.org/10.1016/j.ejogrb.2020.06.054 PMid:32623253
10. Bindra V, Reddy N, Reddy CA, Swetha P, Alapati KV, Nori M. Recurrent perineal scar endometriosis: A case report. Case Rep Womens Health. 2022;36:e00457. https://doi.org/10.1016/j.crwh.2022.e00457 PMid:36281243 PMCid:PMC9587519

11. Barisic GI, Krivokapic ZV, Jovanovic DR. Perineal endometriosis in episiotomy scar with anal sphincter involvement: Report of two cases and review of the literature. Int Urogynecol J Pelvic Floor Dysfunct. 2006;17(6):646-9. https://doi.org/10.1007/s00192-005-0022-5 PMid:16231117
12. Uzuncakmak C, Guldas A, Ozcam H, Dinc K. Scar endometriosis: A case report of this uncommon entity and review of the literature. Case Rep Obstet Gynecol. 2013;2013:386783. https://doi.org/10.1155/2013/386783 PMid:23762683 PMCid:PMC3665185
13. Li J, Shi Y, Zhou C, Lin J. Diagnosis and treatment of perineal endometriosis: Review of 17 cases. Arch Gynecol Obstet. 2015;292(6):1295-9. https://doi.org/10.1007/s00404-015-3756-4 PMid:26041323
14. Tangri MK, Lele P, Bal H, Tewari R, Majhi D. Scar endometriosis: A series of 3 cases. Med J Armed Forces India. 2016;72(Suppl 1):185-8. https://doi.org/10.1016/j.mjafi.2016.07.002 PMid:28050109 PMCid:PMC5192233
15. Blanco RG, Parithivel VS, Shah AK, Gumbs MA, Schein M, Gerst PH. Abdominal wall endometriomas. Am J Surg. 2003;185(6):596-8. https://doi.org/10.1016/S0002-9610(03)00072-2 PMid:12781893
16. Kaplanoglu M, Kaplanoglu DK, Dincer Ata C, Buyukkurt S. Obstetric scar endometriosis: Retrospective study on 19 cases and review of the literature. Int Sch Res Notices. 2014;2014:417042. https://doi.org/10.1155/2014/417042 PMid:27379258 PMCid:PMC4897354
17. Kokuba EM, Sabino NM, Sato H, Aihara AY, Schor E, Ferreira LM. Reconstruction technique for umbilical endometriosis. Int J Gynaecol Obstet. 2006;94(1):37-40. https://doi.org/10.1016/j.ijgo.2006.04.034 PMid:16781715
18. Jain D. Perineal scar endometriosis: A comparison of two cases. BMJ Case Rep. 2013;2013:bcr2013010051. https://doi.org/10.1136/bcr-2013-010051 PMid:23897379 PMCid:PMC3736109
19. Gunes M, Kayikcioglu F, Ozturkoglu E, Haberal A. Incisional endometriosis after cesarean section, episiotomy and other gynecologic procedures. J Obstet Gynaecol Res. 2005;31(5):471-5. https://doi.org/10.1111/j.1447-0756.2005.00322.x PMid:16176520
20. de Paula Andres M, Lopes LA, Baracat EC, Podgaec S. Dienogest in the treatment of endometriosis: Systematic review. Arch Gynecol Obstet. 2015;292(3):523-9. https://doi.org/10.1007/s00404-015-3681-6 PMid:25749349
21. Watanabe M, Kamiyama G, Yamazaki K, et al. Anal endosonography in the diagnosis and management of perianal endometriosis: Report of a case. Surg Today. 2003;33(8):630-2. https://doi.org/10.1007/s00595-003-2545-z PMid:12884104
22. Odobasic A, Pasic A, Iljazovic-Latifagic E, et al. Perineal endometriosis: A case report and review of the literature. Tech Coloproctol. 2010;14(Suppl 1):S25-7. https://doi.org/10.1007/s10151-010-0642-8 PMid:20862505
23. Grimstad FW, Carey E. Periclitoral endometriosis: The dilemma of a chronic disease invading a rare location. J Minim Invasive Gynecol. 2015;22(4):684-6. https://doi.org/10.1016/j.jmig.2015.02.002 PMid:25680686
24. Cinardi N, Franco S, Centonze D, Giannone G. Perineal scar endometriosis ten years after Miles' procedure for rectal cancer: Case report and review of the literature. Int J Surg Case Rep. 2011;2(6):150-3. https://doi.org/10.1016/j.ijscr.2011.04.001 PMid:22096711 PMCid:PMC3199622