



Case report

Case report: Caesarean scar endometriosis – A rare entity

Sigit Purbadi^{*}, Gatot Purwoto, Hariyono Winarto, Kartiwa H. Nuryanto, Laurensia Scovani, Gilbert Elia Sotarduga

Division of Gynecologic Oncology, Department of Obstetrics and Gynecology, Cipto Mangunkusumo Hospital, Jakarta, Indonesia

ARTICLE INFO

Keywords:

Caesarean section
Endometriosis
Post-operative scar

ABSTRACT

Introduction and importance: Caesarean scar endometriosis (CSE) is a rare form of endometriosis due to previous surgical scars from obstetrical and gynecological procedures. The incidence of CSE was 0.08% and quite difficult to diagnose.

Case presentation: A 37-year-old multiparous woman came with intermittent pain in her lower left abdominal region and lump with a bluish color and solid consistency on the left side of the caesarean scar. Ultrasounds findings show a solid mass, measured 45 × 40 × 39 mm with neovascularization color score: 4. In April 2021, we performed mass resection, intraoperatively we found solid mass with no adhesion and infiltration found. Histological examination results confirm external endometriosis from the mass and the fascia was free from endometriosis. The symptoms reported relief after the procedure.

Clinical discussion: Scar endometriosis is largely related to previous abdominal surgery like caesarean section, the mass increase in size during menses and becomes symptomatic. CSE develop ranging 12 months to 21 years and could mimic other hernias or tumor, that's why careful and precise examination is needed. The imaging modality we use in this case was ultrasonography which is the best and most accessible, reliable and cost-effective to diagnose. We performed large surgical excision of the lesion with reconstruction of damaged tissue to prevent recurrence and conversion to malignancy.

Conclusion: Caesarean scar endometriosis should be considered in women of reproductive age with lower abdominal pain and/or mass at the caesarean scar from previous delivery or following obstetric-gynecologic surgery.

1. Introduction

Endometriosis is a sex hormone-dependent gynecological disease where the functional and morphological endometrial tissues are present outside the uterine cavity [1]. Affecting an estimated 89 million women of reproductive age worldwide, endometriosis occurs in 5% to 10% of all women, often resulting in debilitating pain and infertility [2].

Scar endometriosis is a rare form of endometriosis that is usually confused with other surgical or dermatological lesion [3]. Abdominal wall endometriosis (AWE) is one of the most frequent extra pelvic locations, mostly occur due to previous surgical scars from obstetrical and gynecological procedures such as caesarean delivery, hysterectomy, episiotomy, and tubal ligations [4–6].

Caesarean Scar Endometriosis (CSE) is the most commonly reported type of AWE. Nominato et al., said that CS greatly increased the risk of developing AWE due to its pathophysiology where endometrial tissue

from caesarean incision directly implanted to the scar. It is a rare disease, with a reported incidence of 0.03–0.45% CSE may cause long-term discomfort involving cyclic lower abdominal pain [7–9].

2. Case description

A 37-year-old multiparous woman came to our hospital complaining about intermittent pain in her lower left abdominal region. She also complains about pain during her period that keeps getting worse, and the pain was moderate to severe. This complaint was felt for around six years, and she never experienced this symptom before. She had a history of previous caesarean sections twice in 2010 and 2014. There was no previous disease, medication consumption, and history of malignancy in the family recorded from the patient's history. Her daily activities are considered as a moderate activity as a housewife. From the obstetrical history, the caesarean section was performed with Pfannenstiel incision

^{*} Corresponding author.

E-mail address: sigitpurbadi@gmail.com (S. Purbadi).

<https://doi.org/10.1016/j.ijscr.2021.106204>

Received 3 June 2021; Received in revised form 14 July 2021; Accepted 14 July 2021

Available online 16 July 2021

2210-2612/© 2021 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

without any post-operative complication. She consulted the symptoms to a gynecologist several times, but no pathological signs were found, and a pain killer was given to take during her period.

For the last year the pain became greater until March 2021 she found a lump at the left end of the caesarean scar. From the physical examination, the lump was a caesarean scar mass on the skin with a bluish color and solid consistency on the left side of the caesarean scar. The lump size was 4 cm × 4 cm (Fig. 1). The external and internal genitalia were normal. The blood test showed no abnormal findings.

From ultrasound we found a solid mass, the biggest measured size was 45 × 40 × 39 mm with neovascularization color score 4 (Figs. 2 and 3). A differential diagnosis of scar endometriosis or hematoma was made.

In April 2021, based on the examination result mass resection was performed by a gynecologist, including the fascia as the deepest border. Intraoperatively was found solid mass with a diameter of 5 cm, soft consistency, and clear border (Fig. 4). No adhesion and infiltration were found.

Histological examination results confirm external endometriosis from the mass and the fascia was free from endometriosis (Fig. 5). We observed the patient post-operative during routine control for her wound and the patient showed a great improvement. The patient reported a relief of symptoms after the procedure and no evidence of endometriosis was found. No further therapeutic intervention was done to the patient, and it was considered as a complete resection of endometriosis.

Timeline	
Date	Information
2010	1st caesarean section (Pfannenstiel)
2014	2nd caesarean section (Pfannenstiel)
2014–2020	Moderate to severe pain in the lower abdomen especially during periods and keeps getting worse. She consulted this complaint to the gynecologist and was given a pain-killer to take during the period.
March 2021	Found a lump at the left end of the caesarean scar. There was a mass on the skin with a bluish color and solid consistency on the left side of the caesarean scar. The lump size was 4 cm × 4 cm. From the ultrasound, there was a solid mass, with size 45 × 40 × 39 mm, with neovascularization color score: 4.
April 2021	Mass resection, including the fascia as the deepest border. The histological examination confirmed external endometriosis and the fascia was free from endometriosis.

3. Discussion

Scar endometriosis is largely related to previous abdominal surgery, especially caesarean section and early hysterectomy. Abdominal wall endometriosis (AWE) is one of the most frequent extra pelvic locations,



Fig. 1. The mass showed in physical examination.

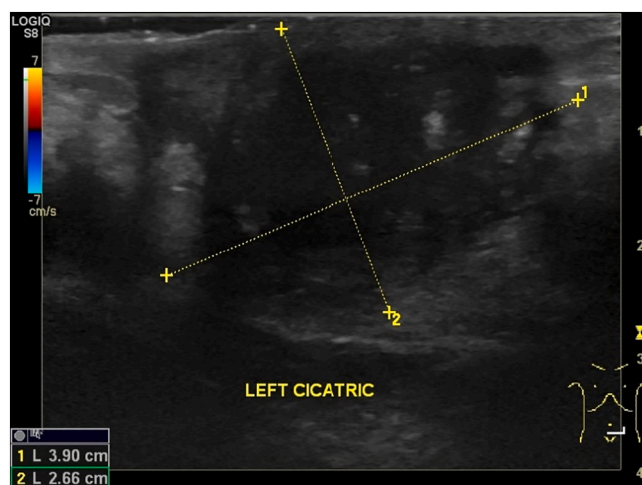


Fig. 2. Ultrasound result showed the size of the mass.

mostly occur due to previous surgical scars from obstetrical and gynecological procedures such as caesarean delivery, hysterectomy, episiotomy, and tubal ligations [4–6]. One study which analyzed 30 years of incisional endometriosis after caesarean section shows that the incidence of scar endometriosis is 0.08% [10]. The frequency of scar endometriosis increases as the high rate of caesarean section and laparoscopy performed in recent years with the incidence of post caesarean section scar quoted to be 1.96% in a recent study [11,12].

We reported an endometriosis case that occurs years after caesarean section [13]. Most studies said that scar endometriosis involves surgical procedures in which the uterus is opened, endometrial tissue gets accidentally implanted into the abdominal incision and it grows to form a mass that increases in size during menses and becomes symptomatic, this is the most plausible theory named direct mechanical implantation [14]. However, this theory cannot explain all cases where few cases of primary cutaneous endometriosis without prior abdominal surgery such as vulva, perineum, groin, umbilicus, and extremities, hence these types of endometriosis could be caused by tubal retrograde spread, genetic and immunologic influences, lymphatic and vascular spread [15]. In our case, the patient did not have prior history of endometriosis or other immunologic disease. From this fact we assume the main cause of endometriosis in this patient was from direct implantation that could be happened during intraperitoneal lavage with sterile water. CSE can develop ranging 12 months to 21 years because it could be remain dormant for several years until it shows symptoms, the common symptoms include cyclical pain, swelling which worsen in the menstrual cycle [3]. CSE can be diagnosed by physical examination and comprehensive history-taking, it presents with mass near previous surgical scar associated with regularly repeating colic pain, which also appeared in our cases [16]. Because abdominal wall endometriosis could mimic incisional and ventral hernias, hematomas, benign or malignant subcutaneous tumors, high awareness and suspicion are required to make diagnosis. We also need to use ultrasonographic examination to prove our suggestion, it is the best and most accessible, reliable, and cost-effective imaging technique to diagnose CSE. The mass will appear hypoechoic and heterogeneous with messy internal echoes [15,17]. Advance imaging like computed tomography and MRI can be an advantage to support our diagnosis. A study said that MRI is a useful modality for presurgical mapping of deep pelvis endometriosis with sensitivity and specificity that could reach 90%–92% and 91%–98%, respectively [18]. In our case, we did not do the advanced imaging because what we got from history taking, physical examination and ultrasound imaging we could conclude that it was a SCE. Some studies reported that we can use fine-needle aspiration cytology (FNAC), instead of its usability of confirming definitive diagnosis, we must be aware

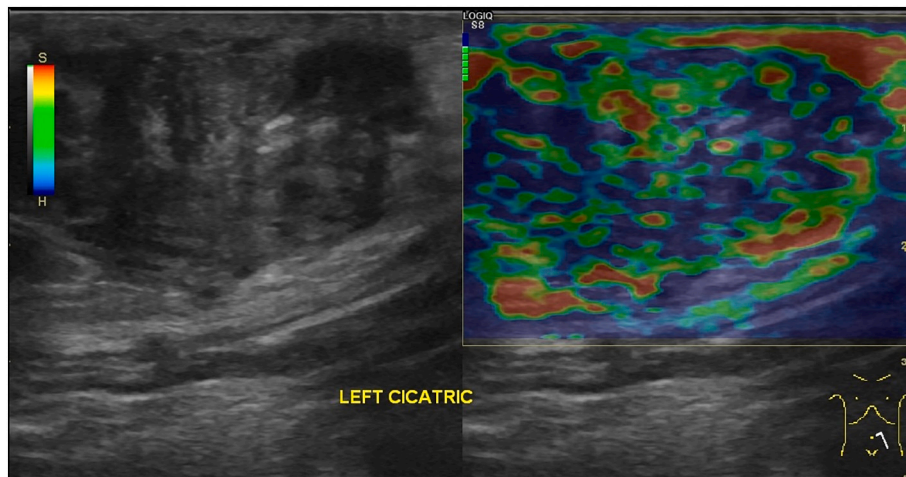


Fig. 3. Ultrasound showed neovascularization from Doppler.



Fig. 4. Macroscopic appearance of endometriosis mass.

about the increasing risk of producing new endometriotic implants at the puncture side [19]. In our case, we did not perform FNAC on this patient.

There are two methods to treat SCE, surgical or non-surgical. For the non-surgical, we could use gonadotropin-releasing hormone analogs which help alleviate clinical symptoms, but this method did not reduce the size and recurrence after the cessation of medication is constant [19]. A surgical procedure like we do in our cases is an accurate treatment of choice of SCE and also for recurrent lesions, as expected, the larger and deeper lesions to the muscle or the fascia are more difficult to

excise completely. For some large lesion cases, complete excision of the lesion may entail a synthetic mesh placement or tissue transfer for closure after resection [20]. Recent studies also suggest the need to clean the abdominal wound with saline solution before closure especially in corner sites to prevent recurrence, careful flushing and irrigation of the adipose layer and fascia layer during closure is critical [21]. From our experience, we suggested large surgical excision of the lesion with a reconstruction of damaged tissue for patients with SCE because based on our experience this type of surgery is effective in preventing recurrence and conversion to malignancy.

4. Conclusion

Caesarean scar endometriosis should be considered in women of child-bearing age with lower abdominal pain and/or mass at the caesarean scar from previous delivery or following obstetric-gynecologic surgery. Both imaging and histopathological examination play important roles in making the diagnosis. Free margin endometriosis excision surgical procedure was the recommended management to excise the lesion completely.

5. Patient's perspective

After I had my second caesarean section, I started to get this pain in my lower abdominal. The pain is felt more significant during my period. At first, I thought this was only a usual menstrual pain. I went to a gynecologist to consult my pain but there was nothing abnormal to be found. After six years, on March 2021 I found a lump in my left side post-caesarean scar. I was worried that it could be a malignant mass even though I never had any history of malignancy in my family. I went to the

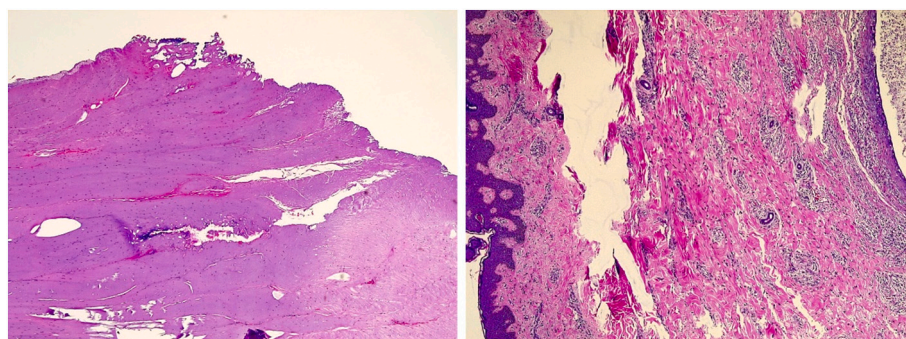


Fig. 5. Histological result from resection. (Left) The base of the fascia showed free from endometriosis. (Right) The skin with external endometriosis.

gynecologist to check the lump. I was then going through further evaluation to confirm the diagnosis with ultrasound, and I was diagnosed with skin endometriosis originated from my caesarean scar. After that, I was observed in the ward and scheduled for a mass resection. The surgery went well, and the histological examination showed the mass was only fixated to the skin and did not invade into the deeper abdominal layer. I was then observed for two days post-operative and then discharged. After the surgery, I felt the pain was significantly decreasing. In my last follow-up, the doctor said that the mass was completely extracted.

Sources of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

This study was reviewed and approved by the Institutional Review Board and Ethical Committee Dr. Cipto Mangunkusumo, a national reference, and teaching hospital. Patient medical records were maintained under applicable medical ethical standards.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Author contribution

Sigit Purbadi: conceptualization, methodology, resources, supervision. Sigit Purbadi, Gilbert Elia Sotarduga, Laurensia Scovani: writing-original draft preparation, investigation, visualization, writing-review and editing. Sigit Purbadi, Gatot Purwoto, Hariyono Winarto, Kartiwa H. Nuryanto: supervision, data curation, editing.

Research registration

None declared.

Guarantor

Sigit Purbadi.

Declaration of competing interest

The authors declare that we have no financial or personal relationship that may have inappropriately influenced us in writing this article.

Acknowledgments

None.

References

- [1] H.E. Williams, S. Barsky, W. Storino, Umbilical endometrioma (silent type), *Arch. Dermatol.* 112 (10) (1976) 1435–1436, <https://doi.org/10.1001/archderm.1976.01630340053014>.
- [2] P. Gupta, P. Gupta, S. Gupta, CASE REPORT scar endometriosis: a case report with literature review, *Acta Med. Iran* 53 (2015).
- [3] C. Douglas, O. Rotimi, Extragenital endometriosis - a clinicopathological review of a Glasgow hospital experience with case illustrations, *J. Obstet. Gynaecol. (Lahore)* 24 (7) (2004) 804–808, <https://doi.org/10.1080/0144361040009568>.
- [4] L. Padmanabhan, R. Mhaskar, A. Mhaskar, Scar endometriosis, *J. Obstet. Gynecol. India* 53 (1) (2003) 59–61.
- [5] R.N. Bhowmick, P. Paul, S. Dutra, B. Roy, Endometriosis of laparotomy scar (a review of 13 cases), *J. Obstet. Gynecol. India* 36 (1) (1986) 130–131.
- [6] S.K. Chatterjee, Scar endometriosis: a clinicopathologic study of 17 cases, *Obstet. Gynecol.* 56 (1) (1980) 81–84.
- [7] J.D. Horton, K.J. DeZee, E.P. Ahnfeldt, M. Wagner, Abdominal wall endometriosis: a surgeon's perspective and review of 445 cases, *Am. J. Surg.* 196 (2008) 207–212, <https://doi.org/10.1016/j.amjsurg.2007.07.035>.
- [8] N.S. Nominato, L.F.V.S. Prates, I. Lauer, J. Morais, L. Maia, S. Geber, Caesarean section greatly increases risk of scar endometriosis, *Eur. J. Obstet. Gynecol. Reprod. Biol.* 152 (1) (2010) 83–85, <https://doi.org/10.1016/j.ejogrb.2010.05.001>.
- [9] P. Reddi Rani, S. Soundararaghavan, P. Rajaram, Endometriosis in abdominal scars - review of 27 cases, *Int. J. Gynecol. Obstet.* 36 (3) (1991) 215–218, [https://doi.org/10.1016/0020-7292\(91\)90716-1](https://doi.org/10.1016/0020-7292(91)90716-1).
- [10] S. Minaglia, D.R. Mishell, C.A. Ballard, Incisional endometriomas after cesarean section: a case series, *J. Reprod. Med.* 52 (7) (2007) 630–634.
- [11] Ö. Aydın, Scar endometriosis - a gynaecologic pathology often presented to the general surgeon rather than the gynaecologist: report of two cases, *Langenbeck's Arch. Surg.* 392 (1) (2007) 105–109, <https://doi.org/10.1007/s00423-006-0107-8>.
- [12] J. Zhang, X. Liu, Clinicopathological features of endometriosis in abdominal wall—clinical analysis of 151 cases, *Clin. Exp. Obstet. Gynecol.* 43 (3) (2016) 379–383.
- [13] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, A. Thoma, et al., The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, *Int. J. Surg.* 1 (84) (2020) 226–230, <https://doi.org/10.1016/j.ijsu.2020.10.034>.
- [14] M. Gunes, F. Kayikcioglu, E. Ozturkoglu, A. Haberal, Incisional endometriosis after cesarean section, episiotomy and other gynecologic procedures, *J. Obstet. Gynaecol. Res.* 31 (5) (2005) 471–475, <https://doi.org/10.1111/j.1447-0756.2005.00322.x>.
- [15] S.C. Ideyi, M. Schein, M. Niazi, P.H. Gerst, Spontaneous endometriosis of the abdominal wall, *Dig. Surg.* 20 (3) (2003) 246–248, <https://doi.org/10.1159/000070392>.
- [16] C. Cöl, E.E. Yilmaz, Cesarean scar endometrioma: case series, *World J. Clin. Cases* 2 (5) (2014) 133–136, <https://doi.org/10.12998/wjcc.v2.i5.133>.
- [17] J.-H.J. Hensen, A.C. Van Breda Vriesman, J.B.C.M. Puylaert, Abdominal Wall endometriosis: clinical presentation and imaging features with emphasis on sonography, *Am. J. Roentgenol.* 186 (3) (2006) 616–620, <https://doi.org/10.2214/AJR.04.1619>.
- [18] K. Kinkel, K.A. Frei, C. Baileysguier, C. Chapron, Diagnosis of endometriosis with imaging: a review, *Eur. Radiol.* 16 (2) (2006) 285–298, <https://doi.org/10.1007/s00330-005-2882-y>.
- [19] F. Cherrabi, M. Moukit, J. Kouach, D.M. Rahali, M. Dehayni, <article-title>Caesarean scar endometriosis: a case report</article-title> <journal-title>Int. J. Reprod. Contracept. Obstet. Gynecol.</journal-title>, *Int. J. Reprod. Contracept. Obstet. Gynecol.* 7 (3) (2018 Feb 27) 1221, <https://doi.org/10.18203/2320-1770.ijrcog20180922>.
- [20] G.K. Patterson, G.B. Winburn, Abdominal wall endometriomas: report of eight cases, *Am. Surg.* 65 (1) (1999) 36–39.
- [21] P. Zhang, Y. Sun, C. Zhang, Y. Yang, L. Zhang, N. Wang, et al., Cesarean scar endometriosis: presentation of 198 cases and literature review, *BMC Womens Health* 19 (1) (2019) 14, <https://doi.org/10.1186/s12905-019-0711-8>.