Rectus abdominis endometriosis: A case report*

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ABSTRACT

Abdominal wall endometriosis is suspected in patients who complain of cyclic tender mass within or adjacent to a caesarean section scar. Ultrasound, magnetic resonance image and computed tomography are helpful tools used to diagnose abdominal endometriosis however histologic examination is required for confirmation. The standard treatment for abdominal wall endometrioma is surgical excision. Proper surgical techniques could prevent abdominal wall endometriosis after uterine surgery. This is a case of a 30 G2P2 (2002) who presented with paraumbilical pain after 2 cesarean sections and previous excision of abdominal wall endometrioma. She underwent excision of the mass and histopath confirmed the presence of endometriosis in the rectus abdominis muscle. Abdominal wall endometrioma is often found in the subcutaneous fatty layer. Its presence in the rectus abdominis is quite rare.

Keywords: abdominal wall endometriosis, rectus abdominis endometriosis

INTRODUCTION

ndometriosis was first described by an Austrian pathologist, Karl Freiherr von Rokitansky in 1860 who referred to the disease as "adenomyoma". The presence of endometrial glands and stroma outside the uterine cavity was then defined by Recklinghausen in 1896. and Sampson described it in detail in 1921. Endometriosis is defined as the growth of ectopic endometrial tissue (glands and stroma) outside the endometrium and myometrium. The true prevalence of endometriosis is uncertain. It is estimated to affect 6-10% of women in the reproductive age group¹. The term endometrioma is given to endometriosis when it forms a well circumscribed mass. Although it is more common in pelvic regions such as the ovaries, pelvic peritoneum, posterior cul-desac, ligaments of the uterus, and rectovaginal septum, endometriosis can also be extrapelvic². It is uncommon to have extrapelvic endometriosis that can affect unusual sites including the urinary tract, gastrointestinal tract, omentum, lymph nodes, hernia sac, umbilicus, extremities, brain and thorax. When this endometrial tissue involves the abdominal wall, usually, it is associated with a history of obstetrical or gynecological procedures such as cesarean delivery, hysterotomy, hysterectomy, and tubal ligation. Interestingly, this phenomenon also has

CASE REPORT

The patient is a 30 G2P2 (2002) who presented with paraumbilical pain. In 2008, the patient underwent primary low transverse cesarean section in another institution secondary to cephalopelvic disproportion. The course of pregnancy, delivery and postpartum period were uneventful. There was no mention of pelvic endometriosis in the caesarean operative technique. In 2010, she noticed a palpable mass superior to the cesarean section scar associated with moderate pain. Both symptoms were present only during the first 2 days of menstruation. She sought consult with the Obstetrician-Gynecologist who performed her cesarean section and surgery was advised. In 2011, she underwent excision of the mass and histopath confirmed the presence of surgical site endometriosis. Operative technique only mentioned the excision of the

occurred after appendectomy, episiotomy, laparoscopic procedures, amniocentesis, inguinal herniorrhaphy and has even been described in patients without any previous surgical interventions^{3,4}. Due to its rarity and the necessity of histological confirmation for diagnosis, it is difficult to estimate the exact incidence of abdominal wall endometriosis. Surgical scar of cesarean section is the most common site of abdominal wall endometriosis with an incidence of 0.07%-0.47%². The incidence of abdominal wall endometriosis is 1.08-2% after hysterotomy³ and 0.5-7% in scars after laparoscopic procedure⁶. Search for local literature revealed one case of rectus abdominis endometriosis without previous surgery⁴.

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mass and failed to describe the exact location of the endometrioma. She still experienced minimal pain in the surgical site during menstruation with no associated palpable mass. She was given continuous combined oral contraceptives (desogestrel + ethinyl estradiol) for 2 years (2011-2013) which resulted in the relief of the cyclical pain. She discontinued the pill with the desire to get pregnant again. In 2014, she delivered via repeat low transverse cesarean section with bilateral tubal ligation in the same institution as the primary surgery. Again the prenatal and postpartum course was uneventful. There was no mention of pelvic endometriosis in the second caesarean operative technique. In 2015, the patient again noticed a palpable mass superior to the cesarean section scar associated with moderate pain during menstruation. She did not seek consult until 2016 when she noticed the pain was present intermittently all throughout the cycle. The mass could also be palpated anytime of the month but was more painful and discrete during menstruation. She was then given Medroxyprogesterone (Depo Trust) 150 mg intramuscularly every 3 weeks for 9 doses but the symptoms persisted despite amenorrhea. Three months prior to present consult, she was shifted to combined oral contraceptives (drospirenone +ethinyl estradiol) but again symptoms persisted.

The patient's past medical history is unremarkable. Her parents are both diagnosed with hypertension. She is a factory worker and has been married for 9 years to a 39-year-old factory worker. She is a non smoker and non alcoholic beverage drinker. Her menarche was at 14 years old with menses occurring at 28-30 days interval lasting for 3-5 days, using 2 moderately soaked pads per day with no dysmenorrhea.

At the time of initial physical examination, the patient was on the day 20 of her cycle. She had a BMI of 20 and stable vital signs. A 6.5 cm midline vertical infraumbilical incision scar was noted with a vaguely palpable, movable and tender mass 1 cm lateral and 2 cm superior to the upper border of the scar. No abdominal wall discoloration was noted. Pelvic exam was normal. Patient was requested to come back during her menses (day 2-5) as well as have a transvaginal with abdominal ultrasound done during her menses.

On day 3 of her menses, a 4x3 cm very tender movable mass was palpated 2 cm lateral and 2 cm superior to the upper border of the scar with no obvious skin discoloration. Pelvic exam was normal. Pelvic and abdominal ultrasound revealed a 3.11 x 1.57 x 2.38 cm well circumscribed heterogenous mass at the subcutaneous layer, 1 cm lateral to the abdominal vertical scar and 3.1cm from the skin. The uterus and ovaries were normal on sonography. (Figures 1 and 2).

Initial impression was abdominal wall endometrioma.

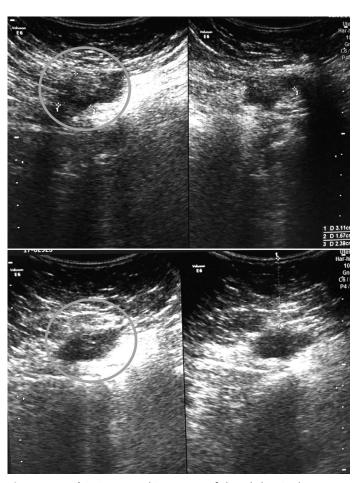
The patient was scheduled for excision of abdominal wall tumor during her next menses. Intraoperatively, a 6.0 x 5.0 x 2.0 cm in diameter mass that was located 2 cm superior and lateral to the incision was reached within the right rectus abdominis muscle. (Figure 3) The lesion was ex–cised together with a 1 cm margin surrounding intact muscular tissue. Examination of the pelvic cavity did not reveal any evidence of pelvic endometriosis. The patient was referred intraoperatively to surgery for abdominal wall mesh repair (Figure 4). The mass was firm in consistency and on cut section; there was note of tar like fluid and presence of blood lakes. (Figure 5) Postoperative course of the patient was unremarkable.

Histopathology confirmed endometrioma with all surgical margins of resection negative for endometriosis. (Figure 6).

Final impression was rectus abdominis endometrioma.

At present, one year after her surgery, there has been no recurrence of abdominal pain or mass associated with

menstruation.



Figures 1 and 2. Sonographic Report of the Abdominal Mass

Gynecology Ultrasound Report (TAS)

I. Uterine Corpus: 5.10 x 4.51 x 4.20 cm LxWxH Retroverted

II. Cervix: 2.19 x 2.48 x 1.84 cm LxWxH

Nabothian Cyst: None

Description/Abnormalities: None

III. Endometrium: 0.72 cm Isoechoic

Compatible with Proliferative phase of menstrual cycle

IV. Adnexae Right Ovary: 2.15 x 1.16 x 2.35 cm LxWxH Vol = 3.07

Located: Lateral Follicle: Present Abnormalities: None

Left Ovary: 2.69 x 1.91 x 1.42 cm LxWxH Vol = 3.82

Located: Lateral Follicle: Present Abnormalities: None

V. Cul de Sac: No free fluid in the posterior cul de sac

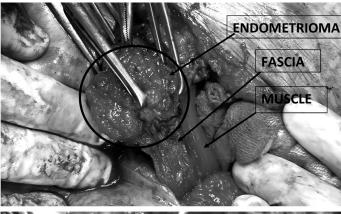
VI. Color Doppler Study: N/A

VII. Others: There is a 3.11 x 1.57 x 2.38 cm, well circumscribed heterogenous mass at the subcutaneous layer, 1 cm lateral from the abdominal vertical scar (laterality), and 3.1 cm from the skin (depth).

DIAGNOSIS: Normal sized uterus with intact endometrium

Normal cervix Normal ovaries

Abdominal mass (Deep subcutaneous layer)



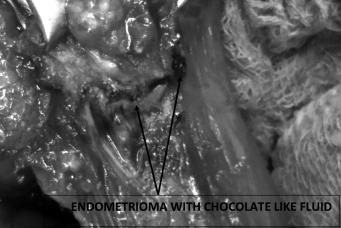


Figure 3. Intraoperative Pictures

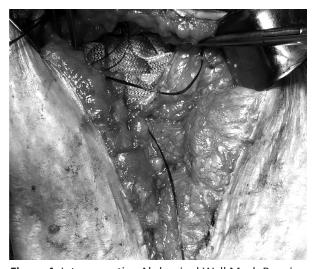


Figure 4. Intraoperative Abdominal Wall Mesh Repair

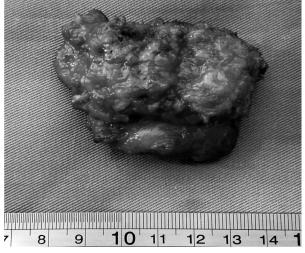


Figure 5. Abdominal Wall Endometrioma

Histopathology Report: Endometriosis Negative Endometriosis. All surgical margins of resection

The specimen labeled is ABDOMINAL WALL ENDOMETRIOMA consists of previously opened muscle tissue with cystic spaces embedded within and contains reddish-brown fluid, measuring $5.0 \times 4.0 \times 2.5$ cm. Cut section show an irregularly-shaped, cream white to tan, solid area measuring $2.5 \times 2.0 \times 1.0$ cm.

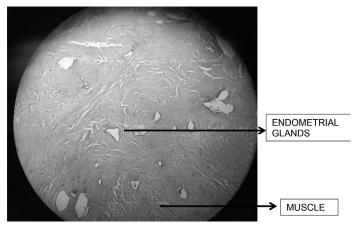


Figure 6. Histopathology report

CASE DISCUSSION

Studies on risk factors for abdominal wall endometriosis are scarce. Caesarean section is the most common procedure related to abdominal wall endometriosis being present in 81% of cases⁷. This was likewise present in the index patient who underwent 2 cesarean sections. Other risk factors include an early hysterotomy in pregnancy, increased menstrual flow, elevated body mass index, and alcohol consumption^{7,8}. The reason for higher incidence after hysterotomy is due to the early decidua having more pluripotential capabilities and can result in cellular replication producing endometrioma³. Interestingly, abdominal wall endometriosis was identified in the 9% of patients with no prior history of abdominal surgeries⁷.

The average time between prior abdominal surgery and the first presentation of abdominal wall endometriosis is about 3-7 years with the range being from 3 months to 10 years ^{3,5,7}. Due to this significant delay, it is important to maintain a high clinical suspicion when the symptoms are suggestive of abdominal wall endometriosis. A painful palpable mass near an incisional scar that may or may not be related to the menstrual cycle is pathognomonic for abdominal wall endometriosis⁸. Occasionally, abdominal wall endometriomas may present as a painless but gradually enlarging mass. *The patient initially presented with cyclical surgical site tender mass 2 years after her first*

cesarean section. Eventually the mass and pain was noted all throughout the cycle, being more pronounced during menstruation.

The pathogenesis of abdominal wall endometriosis is not yet fully understood and might be best explained by iatrogenic direct implantation theory². In this theory, there is direct inoculation of endometrial tissue into the surgical wound during the surgery. The ectopic endometrium is embedded in the subcutaneous fatty layer and responds to estrogen stimulation. Rectus abdominis endometrioma, wherein the endometrioma is confined only within the rectus abdominis muscle, is quite rare and was first reported in 1993. Another possible theory in the development of abdominal wall endometrioma is the theory of vascular spread⁹. According to this theory, endometrial cells reach extragenital regions through blood vessels or the lymphatic system resulting in endometriotic foci. Both theories could be applied to the index patient. The endometrioma could have developed from direct inoculation after her first cesarean section and the initial excision was incomplete thus the symptoms persisted. But since the mass excised from the second excision was not at the surgical site, the development of the rectus muscle endometriosis can also be explained by the vascular theory.

It is difficult to establish whether the rectus abdominis endometriosis of the patient is a recurrence after the first excision or a new endometriosis occurring after the second caesarean section. The recurrence of abdominal wall endometriosis at the same location after surgical excision is infrequent. Its incidence is 0.5 % to as high as 29 %7. The simultaneous occurrence of pelvic endometriosis with scar endometriosis is uncommon.

Endometriosis of the abdominal wall maybe difficult to diagnose; it is often mistaken, both clinically and with diagnostic imaging, for other conditions such as a suturegranuloma, an incisional hernia, fibrosis, fat necrosis, abscess and primary or metastatic malignant lesions². Ultrasonography is the most frequently utilized diagnostic modality technique for diagnosing abdominal wall endometriosis. Sonologically, the masses appear solid, hypoechoic and contain internal vascularity on doppler examination¹⁰. These sonographic findings are non-specific but when coupled with history of surgical intervention and physical findings, an appropriate diagnosis can be made. The major role of computed tomography and magnetic resonance imaging is to depict the extent of the disease preoperatively. Fine needle aspiration cytology may also be used in accessible abdominal masses to aid in the diagnosis. Sonographic findings in the index patient revealed that the abdominal mass was in the subcutaneous layer where in fact it was localized in the rectus abdominis, implicating the limitations of sonography.

Wide surgical excision (with margins of at least 1

cm) is the definitive treatment of choice for abdominal wall endometrioma⁹. This is to prevent recurrence and risk for conversion to malignancy, although this phenomenon is very rare (less than 1%). Abdominoplasty and reconstruction with or without polypropylene mesh should be considered if a defect in the abdominal wall occurs, which may be caused by the wide excision of the muscular layer of the abdomen, as what was done to the index patient.

Medical therapy can be used to relieve the clinical symptoms of abdominal wall endometriosis and often involves hormone suppression in the form of progestogens, combined oral contraceptives, danazol and gonadotropin releasing hormone agonist. The success rate of medical therapy has been reported to be low, offering only temporary relief of symptoms followed by recurrence after cessation of the drug¹⁰.

Proper surgical techniques may prevent the

abdominal wall after any uterine surgery and reduce the risk of endometrial cellular spread. These techniques include (1) isolated individual gauzes for endometriosis lesions, (2) careful irrigation, (3) use of wound edge protectors (4) usage of different needles to repair the abdominal wall and (5) closure of the anterior peritoneum¹¹.

CONCLUSION

Painful swelling in the abdominal scar in the background of previous gynecological or obstetrical surgery can be confused with surgical conditions and scar endometriosis. A high index of suspicion is necessary to clinch the diagnosis. Preoperative evaluation can be done using imaging techniques and fine needle aspiration cytology. Wide excision with free margins is considered the best treatment in localized masses since medical treatment is not helpful.

REFERENCES

- Clinical Practice Guidelines: ENDOMETRIOSIS. Philippine Society of Reproductive Endocrinology and Infertility (2014).
- Tahere Khamechian, Javad Alizargar, Tahere Mazoochi. 5-Year data analysis of patients following abdominal wall endometrioma surgery. BMC Women's Health Journal. 2014 Dec. Avaialable from: https://bmcwomenshealth.biomedcentral.com/articles/10.1186/ s12905-014-0151-4.
- Prariksha Gupta and Sangeeta Gupta. Scar Endometriosis: a Case Report with Literature Review. Department of Gynecology and Obstetrics, Post Graduate Institute of Medical Sciences and Research ESIC, New Delhi, India. 2014 Jan. page 793-795. Available from: https://www.ncbi.nlm.nih.gov/pubmed/26749239.
- 4. Charisse Espina-Tan and Christine Joy Ouano Rectus Abdominis endometriosis without previous surgery: a case report. *Philippine Journal Of Obstetrics and Gynecology*. June 2010; 34:76-81.
- Tihomir VukšiT, Pejana Rastovic and Vedran Dragisic. Abdominal Wall Endometrioma after Laparoscopic Operation of Uterine Endometriosis. Hindawi Publishing Corporation. 2016 April. Available from: https://www.hindawi.com/journals/ cris/2016/5843179/
- 6. S. Gonzales-Fernandez, S. Sabra, M. Oltra et. Al,. Case Report of Recurrent Abdominal Wall Endometrioma at the Same Location after Nine Years of Its First Excision. Barcelona, Spain: Journal of Fertilization: In vitro - IVF-Worldwide, Reproductive Medicine, Genetics & Stem Cell Biology. 2017 Feb. Available from: https:// www.omicsonline.org/open-access/case-report-of-recurrentabdominal-wall-endometrioma-at-the-samelocation-after-nineyears-of-its-first-excision-2375-4508-1000198.
- Eun Mee Oh, Won-Suk Lee, Jin Mo Kang et al,. A Surgeon's Perspective of Abdominal Wall Endometriosis at a Caesarean

- Section Incision: Nine Cases in a Single Institution. Hindawi Publishing Corporation. 2014 July. Available from: https://www.hindawi.com/journals/srp/2014/765372/
- Hatice Karaman, Feridun Bulut and Aysel Ozaslamaci. Endometriosis externa within the rectus abdominis muscle. Kayseri, Turkey: Kayseri Training and Research Hospital. Volume 30, Pages 165-168. 2014 April.
- Jan-Hein J. Hensen, Adriaan C. Van Breda Vriesman and Julien B. Puylaert. Abdominal Wall Endometriosis: Clinical Presentation and Imaging Features with Emphasis on Sonography. Netherlands: A Century of Medical Imaging. 2005 Feb.
- Christos Nissotakis, Efstratios Zouros, Kyriakos Revelos MD. Et al., Abdominal wall Wall Endometrioma: A Case Report and Review of the Literature. AORN Journal, Volume 91, pages 730-744. 2010 June.
- Imane Khachani, Abdelhai Filali Adib and Rachid Bezad. Cesarean Scar Endometriosis: An Uncommon Surgical Complication on the Rise? Case Report and Literature Review. Rabat, Morocco: Hindawi Case Reports in Obstetrics and Gynecology . 2017 Feb. available from: https://www.hindawi.com/journals/criog/2017/8062924/
- Murat Bozkurt, Said Cil and Dugyu Kara Bozkurt. Intramuscular Abdominal Wall Endometriosis Treated by Ultrasound-Guided Ethanol Injection. Kars, Turkey: Kafkas University School of Medicine, Department of Obstetrics and Gynecology. 2013 Oct. available from: https://www.ncbi.nlm.nih.gov/pmc/articles/ PMC4317157/
- 13. Gianpaolo Carrafiello, Federico Fontana, Carlo Pellegrino et. Al., Radiofrequency Ablation of Abdominal Wall Endometrioma. Volume 32, pages 1300-1303. Available from https://www.ncbi.nlm.nih.gov/pubmed/19184197