CASE REPORT

Uterus couvelaire after caesarean section: A challenging case report

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ABSTRACT

Objectives: To discuss the discovery of uterine couvelaire events after the cesarean section without accompanying placental abruption.
Case Report: Uterus Couvelaire is a rare occurrence. The incidence of this case is difficult to ascertain and its estimated incidence is as much as 20% and others' estimatation is as low as 5%. It occurs mainly due to complications from placental abruption. When a vascular injury occurs in the placenta, it causes bleeding that infiltrates the wall of the uterus. This case is usually diagnosed accidentally because it is diagnosed only by direct visualization or biopsy. In this case, uterine couvelaire was found in a woman after a cesarean section that had been performed previously. Uterine couvelaire events are usually seen due to complications from placental abruption, but in this case, there was none.
Conclusion: It was not known for sure what caused the emergence of the uterine couvelaire in this case. Hysterectomy was performed in this case due to the patient's unstable hemodynamic state.

Keywords: Uterus couvelaire; cesarean section; hysterectomy; childbirth complications; maternal health

ABSTRAK

Tujuan: Untuk membahas ditemukannya kejadian *uterus couvelaire* setelah tindakan operasi sesar dan tanpa disertai dengan abrupsio plasenta.
Laporan Kasus: *Uterus couvelaire* merupakan kejadian yang jarang terjadi. Insiden kasus ini sulit untuk dipastikan dan diperkirakan kejadiannya terjadi sebanyak 20% dan yang lain memperkirakan serendah 5%. Kejadiannya terutama akibat komplikasi dari abrupsio plasenta. Ketika terjadi cedera vaskular di dalam plasenta menyebabkan perdarahan yang menginfiltasi dinding dari uterus. Kasus ini biasanya terdiagnosis secara tidak sengaja karena didiagnosis hanya dengan visualisasi secara langsung atau biopsi. Pada kasus ini ditemukan kasus *uterus couvelaire* pada seorang wanita setelah sebelumnya dilakukan operasi sesar. Kejadian *uterus couvelaire* biasanya ditemukan akibat komplikasi dari abrupsio plasenta, namun pada kasus ini tidak ada.
Simpulan: Tidak diketahui pasti apa yang sebabkan timbulnya *uterus couvelaire* pada kasus ini. Histerektomi dilakukan pada kasus ini karena keadaan hemodinamik pasien yang tidak stabil.

Kata kunci: *Uterus couvelaire*; sectio caesarea; histerektomi; komplikasi persalinan; kesehatan ibu

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INTRODUCTION

Uterus couvelaire, also known as uteroplacental apoplexy, is a condition that results from complications from placental abruption.[1] It was first described by a French obstetrician named Alexandre Couvelaire in 1911.[2] The incidence of this case is difficult to ascertain, and the estimated incidence is between 5% and 20%.[1] The profile of this case is the extravasation of extensive blood into the uterine muscle and down the surrounding tissue.[3,4] Distinctive appearance is bluish or purple, speckled by ecchymosis.[1] Thereafter effusion of blood are also visible beneath the tubal serosa, between the leaves of the broad ligament, in the substance of the ovaries, and free in the peritoneal cavity.[5]

The pathophysiology of this case is bleeding in the layer between the decidua-placenta, which then develops and infiltrates into the uterine wall. Characteristically it shows the appearance of ecchymosis on the surface of the uterine serosa. Sometimes bleeding can also occur between large layers of ligaments and infiltrating the uterine wall muscles.[6,7] Sometimes, infiltration can also reach the peritoneal cavity.[8] The resulting effect causes contraction of the myometrium or uterine muscle to become weak and even rupture due to an increase in intrauterine pressure.

Although the etiology is unknown, the uterine couvelaire is associated with placental abruption, placenta previa, coagulopathy, pre-eclampsia, uterine rupture, and amniotic fluid embolism.[2,9,10] The way to make a diagnosis of this case is by direct visual inspection of the uterus or by biopsy or both.[1,10-12] Because of the reason for the diagnosis of uterine couvelaire diagnosis, it can only be identified directly by direct inspection or biopsy, so the incidence of this case is not widely reported and is minimal in the literature search.[4,11]

The management of these cases is usually done immediately conservatively, and hysterectomy is usually unnecessary.[1,6,8,13,14] The uterine couvelaire does not affect the ability of the uterus to contract and decompress, allowing narrowing of the arteries to reach a state of hemostasis. Hysterectomy can be indicated as an act of saving the soul if the process of hemostasis is not achieved so that it can cause disseminated intravascular coagulation.[12]

CASE REPORT

A 37-year-old woman, G2P1A0, on April 28, 2020, came with a referral from a local midwife because of a history of premature rupture of membranes in the last 24 hours and parturition. While in the hospital, the patient was observed and tried to do a normal delivery, but finally the patient was delivered by section caesarea due to indications of fetal distress. After surgery, the woman's condition was stable and treated in a standard room. Then, two days later, on April 30, 2020 the patient complained of tightness, abdominal pain and so forth. Physical examination was immediately carried out when the vital signs obtained blood pressure of 100/70 mmHg, pulse 136x/m, breath rate 46x/m, temperature 36.6ºC, and 99% SpO2 by administering oxygen via face mask. Other physical and obstetric examination revealed both conjunctival anemias, abdominal tenderness, active vaginal bleeding ± 200 cc, urine output only ± 5 cc (anuria). Investigations in the form of routine blood obtained patient Hb levels of 2.8 g/dL, hematocrit 8.7 g/dL, platelets 215,000/uL and leukocytes 21,900/uL. Immediately the patient was transfused with 4 units of PRC along with other resuscitation fluids in the form of NaCl liquid as much as 1500 cc. Immediate laparotomy surgery with suspicion of intraabdominal bleeding was planned for the following day.

On May 1 2020 the patient was immediately operated for laparotomy under general anesthesia because of a suspected intraabdominal hemorrhage. At the time of laparotomy, when the abdominal cavity was opened, it was found that the size of the uterus was of eight months gestational age. The hematoma in the uterine wall was bluish and uterine contraction was not palpated (Figure 1).

Figure 1. Uterine size during exploratory laparotomy

Then it was decided to do a hysterectomy because of hemodynamic instability (Figure 2). Uterine tissue biopsy was also performed during hysterectomy and then the tissue was taken for anatomic pathology examination. After surgery, the patient was transferred

to the HCU (High Intensive Care) room for 1 day until May 2, 2020. During treatment in the HCU room, the patient's condition was stable so that she was moved to an ordinary ward. When treated in a standard room, patients only complained of pain at the post-operative site and some nausea, no significant complaints or emergencies were found. Routine blood examination was done on May 3, 2020, revealing Hb of 7.7 g/dL, hematocrit 23.7 g/dL, platelets 176,000/uL and leukocytes 11,500/uL.

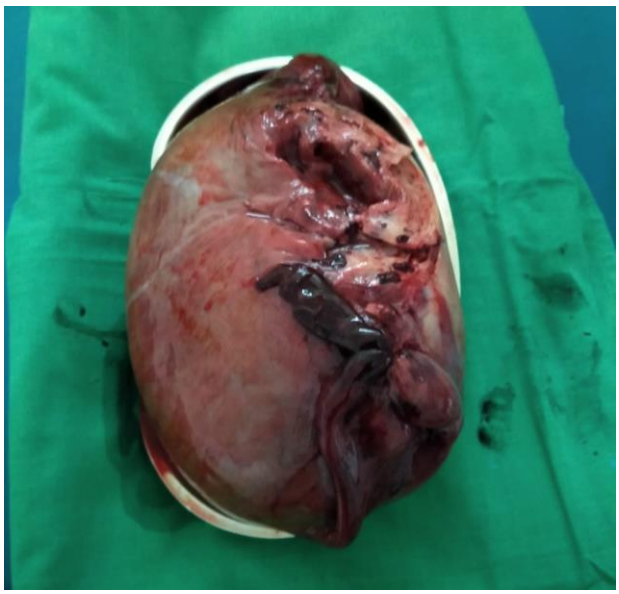

Figure 2. The uterus after hysterectomy

The patient was treated in a ward room until she was allowed to go home on May 5, 2020 with the patient's last vital signs as follows: blood pressure of 120/80 mmHg, pulse 80x/m, breathing rate 21x/m and temperature 36.7ºC. On the pati15ent's discharge date, the histopathological results from the biopsy were released. The conclusion of the results of the specimen obtained was uterine couvelaire. A week after the patient returned from treatment at the hospital, the patient was examined at an obstetric clinic. At that time, the patient was in a stable condition and no complaints were found.

DISCUSSION

Uterine or uteroplacental couvelaire is a condition in which there is bleeding in the uterine wall. Most of these cases occur because of placental abruption. This case was first described in 1911 by a French obstetrician named Alexandre Couvelaire. The incidence of this case is challenging to ascertain because the diagnosis is found accidentally through direct examination or biopsy. Uterus couvelaire, in addition to placental abruption, is also associated with cases of placenta previa, coagulopathy, pre-eclampsia, uterine rupture and amniotic fluid embolism. In the case of uterine couvelaire that we had found, no placental abruption or other comorbidities were found. This case was like the one reported by Kori et al. who wrote that they found a case of uterine couvelaire without considering the presence of placental abruption.[6] It was not known exactly what had caused this uterine couvelaire, because there was no placental abruption, but the incidence occurred several days after cesarean section. When the cesarean section did not find any signs of uterine couvelaire either, so it was likely due to iatrogenic injury to the uterine wall after the section which had caused extravasation of blood. However, there were no literatures or case reports of uterine couvelaire events after cesarean section. In one case reported by Osial et al., they had a uterine couvelaire after curating and dilating, but not because of a cesarean section.[1]

Management is usually conservative because there will be a natural resolution so that the condition will return to normal. Management of the antenatal and postpartum hemorrhage plays a key role, on one side in the control of the bleeding with blood transfusion and reduce fetal and maternal morbidity and mortality.[15] However, if hemostasis does not occur, hysterectomy needs to be done as an act of saving lives. In this case, it was decided that a hysterectomy was performed because of the patient’s unstable hemo-dynamic and also the consideration of the previous patient had performed a cesarean section with the incision line located in the lower uterine segment by the treating doctor. Similar to this case, in the cases reported by Habek et al. and Osial et al. in 2013, they also performed a hysterectomy because of unstable hemodynamic conditions.[2,11]

CONCLUSION

The occurrence of uterine couvelaire is found mostly in cases of placental abruption. However, in this case, the abruption case was absent. Cesarean section was performed unconsciously, followed by the emergence of the uterus couvelaire. Then, hysterectomy was performed to maintain the patient's hemodynamic stability.

REFERENCES

1. Uwagbai ON, Wittich AC. A 30-year-old female found to have a couvelaire uterus with placenta

accreta during planned cesarean delivery. Mil Med. 2017;182(3):e1877-e9. doi: 10.7205/MILMED-D-16-00146.
2. Osial P, Osial P, Atkinson AL, et al. An unreported etiology for couvelaire uterus of case presentation. 2013;2(4):244–8.
3. Mani M, Nidhi A. Couvelaire uterus with concealed abruptio placenta?: A case report. International Journal of Clinical Obstetrics and Gynaecology 2018;2(5):170–1.
4. Senkaya AR, Umut F, Ileri A, et al. Maternal and neonatal outcomes of couvelaire uterus. Med Sci Int Med J Matern. 2021;10(3):775–8.
5. Cunningham FG, Leveno KJ, Bloom SL, Dashe JS, Hoffman BL, Casey BM, et al., editors. Obstetrical Complications. In: William Obstetrics 25th Edition. 25th ed. New York: Mc Graw Hill Education; 2018. p. 771.
6. Kori S, Mudanur SR, Patil N, et al.. Couvelaire uterus without placental abruption: A rare case report. J Evol Med Dent Sci. 2014; 3(38):9787–9.
7. N. Sunanda N, Sruthi T, Sheela SR. Feto-maternal outcome in patients with couvelaire uterus: A 3 year study in a tertiary care hospital in rural Karnataka, India. Int J Reprod Contraception, Obstet Gynecol. 2018;7(2):503. doi: 10.18203/2320-1770.ijrcog20170162.
8. Rathi M, Rathi SK, Purohit M, Pathak A. Couvelaire uterus [Internet]. Case Reports; 2014 [update 2014; cited 2014 Mar 31]. Available from: http://casereports.bmj.com/cgi/doi/10.1136/bcr-2014-204211.
9. Ting JCS, Wong XS, Yong SL, Kang M. A “miracle” pregnancy outcome of severe placental abruption. Hormone Molecular Biology and Clinical Investigation. 2019;38(3):20180059. doi: 10.1515/hmbci-2018-0059.
10. Cipullo LMA, Poto GE, Zullo F, et al. Late complication of a couvelaire uterus?: Case report and literature review. Journal of Pregnancy and Child Health 2019;6(2):6–7. doi: 10.4172/2376-127X.1000407
11. Braila AD, Krastev BM, Mihai-Zamfir E, et al. Uteroplacental apoplexy associated with invasive cervical neoplasm. Rom J Morphol Embryol. 2017;58(4):1465-70. PMID: 29556642.
12. Ming GS, Lee WKR, Tan SQ. An unusual case of placenta abruption leading to couvelaire uterus in a previable pregnancy. J Med Cases. 2020;11(4):103–5. doi: 10.14740/jmc3462.
13. Grohs MA, Kamoune J, Watfeh R, et al. Couvelaire uterus with placental abruption: A striking picture but a normal function. Int. J. of Adv. Res. 2018;6(8): 1028-30. doi:10.21474/IJAR01/7608.
14. Mahendra G, Pukale RS, Vijayalakshmi S. Couvelaire uterus - A case report. Int Arch Integr Med. 2015;2(3):142–5.
15. Cobec IM, Rempen A. Couvelaire-Uterus: literature review and case report. Rom J Mil Med. 2021;124:244–6.