Pregnancy Complicated by Cervical Varix and Low-Lying Placenta: A Case Report

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Abstract: We present a case of cervical varix and low-lying placenta. A cesarean section was performed because of the risk of bleeding with vaginal delivery; hemostasis was achieved using Z sutures at the bleeding points. After delivery, the cervical varix decreased dramatically in size. It is important to recognize the clinical features and available treatments for cervical varix.

Keywords: cervical varix, low-lying placenta, hemorrhage
Introduction
Although vulvar varices complicate approximately 15% of all pregnancies and rarely cause bleeding during pregnancy and delivery, a cervical varix, which is rare, can cause massive obstetric hemorrhage. Its diagnosis, preferable mode of delivery, and prognosis have not been established. We present a case of a cervical varix with a low lying placenta.

Case Report
A 40-year-old Japanese woman (gravida 2, para 1) presented at our hospital at 18 weeks’ gestation for prenatal care. At 22 weeks’ gestation, transvaginal ultrasonography revealed a low lying placenta; the placenta was located on the lower side of the posterior uterine wall, with its lower edge near the internal cervical os. An ultrasound performed at 34 weeks revealed that the cervical tissue had been replaced with a dilated venous plexus with abundant blood flow (Fig. 1). We diagnosed this condition as a large cervical varix. However, we found no specific abnormal findings with the speculum examination. Vaginal bleeding did not occur during her pregnancy. A vaginal delivery was extremely likely to cause catastrophic bleeding; therefore, we performed a planned cesarean section at 37 weeks, yielding a healthy female infant (3,345 g; Apgar score 8/10 (1/5 min)). The placenta was easily separated manually. After placental separation, inspection of the cervical lumen and the lower segment lumen revealed dilated blood vessels protruding into the lumen. Some vessels ruptured with resultant active bleeding, which required several hemostatic Z-sutures (Fig. 2). The estimated blood loss was 3,610 mL; 4 units of packed red blood cells were transfused. The size of the cervical varix decreased dramatically postpartum (Fig. 3). A varix was not present in the cervix two months postpartum.

Comments
We describe a pregnant woman with cervical varices in whom a cesarean section was performed. Even with the cesarean section, active bleeding ensued, which required a blood transfusion and hemostatic sutures. Although the etiology of cervical varices during pregnancy is unclear, a lower placental location, a placenta previa, and a low-lying placenta are considered to be culprits. A lower placental location increases the blood flow to the cervix, leading to the development of cervical venous dilatation, and eventually cervical varices. Hormonal dynamic changes, increased abdominal pressure, and the enlarged uterus that is a result of pregnancy will compress the inferior vena cava and pelvic veins, thus also accelerating varix formation. Although this is rare occurrence, maternal exposure to diethylstilbestrol (DES) in utero causes vascular malformations of pelvic organs in their daughters, which has also been considered to be causative for this disorder.1,2

Figure 1. Transvaginal ultrasound image at 34 weeks of gestation shows severely dilated tortuous blood vessels replacing most of the normal cervical tissue (B) power Doppler.

Figure 2. Photo during operation. Note: Arrow head shows cervical varix with active bleeding.

Figure 3. Transvaginal ultrasound image of the uterus 2 days postpartum (A) and 2 months postpartum (B).
Table 1. Summary of published cases of cervical varices in pregnancy.

| Reference             | Maternal age (years) | G/P | GA at diagnosis | Bleeding during pregnancy | Treatment                          | Placental location | Mode of delivery | GA at delivery | Blood loss (mL) | Birth weight (g) | Apgar score 1 min/5 min |
|-----------------------|----------------------|-----|-----------------|---------------------------|-----------------------------------|-------------------|-----------------|----------------|----------------|--------------------|--------------------------|
| Follem et al²         | 31                   | 3/3 | 22              | Yes                       | Packing, bed rest                 | NS                | CS              | 35.5           | NS             | NS                 | NS                       |
|                       | 32                   | 4/1 | 13              | No                        | Cerclage, bed rest                | NS                | CS              | 31             | NS             | 1700               | NS                       |
| Fleming and Anderson¹ | 32                   | 2/0 | 28              | Yes                       | Bed rest                          | NS                | TVD             | Term           | NS             | NS                 | NS                       |
| Hurton et al⁶         | 26                   | 2/1 | 25              | No                        | Cerclage                          | NS                | CS              | 36.5           | NS             | 3239               | NS                       |
| Yoshimura et al⁷      | 33                   | 2/0 | 33              | Yes                       | Tocolytics, hysterectomy          | Not low           | CS              | 34             | 5,000          | NS                 | 7/8                      |
| Kusanovic et al⁶      | 34                   | 1/0 | 18              | Yes                       | Tocolytics, bed rest              | Previa            | CS              | 27             | 920            | 1,090/920         | 5/7                      |
| Kumazawa et al⁵       | 30                   | 1/0 | 27              | Yes                       | Tocolytics, packing, blood        | Previa            | CS              | 32             | 1,814          | 1,655              | 6/8                      |
| Sammour et al⁴        | 36                   | 1/0 | 23              | No                        | Bed rest                          | Low lying         | CS              | 37             | 1,000          | 3,695              | 8/10                     |
| Sukur et al³          | 40                   | 2/1 | 37              | Yes                       | No                                | Previa            | CS              | 37             | 1,500          | 2,930              | NS                       |
| Present case          | 40                   | 2/1 | 34              | No                        | No                                | Low lying         | CS              | 37             | 3,610          | 3,345              | 8/10                     |

**Abbreviations:** CS, cesarean section; GA, gestational age; NS, not specified; TVD, transvaginal delivery.
While vulvar varices during pregnancy are frequently observed and they rarely cause clinically significant bleeding, cervical varices can cause massive obstetric hemorrhage. A PubMed search yielded only 10 case reports describing cervical varices. In seven of 11 cases (including our case), prenatal bleeding occurred and required termination of pregnancy; in five cases abnormal placental location was recognized (Table 1). Interestingly, two of 10 previously reported cases were from Japan, and the present case was also a Japanese woman. In our case, gray-scale with color Doppler ultrasound detected this abnormality. In the last decade, transvaginal color Doppler ultrasound has been employed in Japan and it is becoming a general-purpose imaging modality in obstetrics. This may be the reason why three out of 11 cases have been reported from Japan. In addition, without the employment of color Doppler, the disorder may remain unrecognized and the cervical variceal rupture could be an undiagnosed factor in cases of massive perinatal hemorrhage. Thus, the incidence of cervical varicosities might be higher than the expected level calculated from earlier case reports.

Due to DES exposure, the placement of a cervical cerclage was performed in two cases. Although bleeding did not occur during these pregnancies, hemorrhagic risk from a cerclage suture does exist. Thus, this surgical option remains controversial.

The cervical varices in the present case ruptured with significant hemorrhage, despite the cesarean delivery. We extended the uterine incision to the cervix and repaired the varices under direct vision. As a consequence, we achieved hemostasis. A hysterectomy or transarterial embolization was avoided. We assume that the cervical varix can be identified by a transvaginal ultrasound, with the awareness of its possible coexistence with abnormal placenta; thus, a management strategy can be developed to prevent postpartum hemorrhage.

Author Contributions
Conceived and designed the experiments: MM, HT, DT. Analyzed the data: YK, MT, MM. Wrote the first draft of the manuscript: YK, DT. Contributed to the writing of the manuscript: YK, MT, MM, DT. Agree with manuscript results and conclusions: YK, MT, MM, DT. Jointly developed the structure and arguments for the paper: MM, HT, DT. Made critical revisions and approved final version: TS, MK, OI. All authors reviewed and approved of the final manuscript.

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