Journal of Rare Diseases

CASE REPORT

Open Access

Rescue cervical cerclage in twin pregnancy with spontaneous cervical dilatation at 8-week gestation

Gaurav Sharma[1*], Samir Pradhan[2], Sara Zaidi[3], Rinoy Shreedharan[1], Vipin Chandra[4] and Nihar Ranjan Bhoi[4]

Abstract

Premature spontaneous cervical dilatation can happen among pregnant women leading to miscarriage, preterm labor, or premature delivery. We present a case of twin pregnancy with spontaneous cervical dilatation and membranes bulging into the vagina at 8-week gestation. A 34-year-old primigravida presented to our clinic with complaints of cramping pain in the lower abdomen. Her per speculum pelvic examination showed evidence of dilated cervical os and amniotic membranes bulging through the cervix into the vagina. Her sonogram suggested a dichorionic diamniotic pregnancy of 8-week gestation. Her TVS examination revealed that the internal os was dilated. Both fetuses were alive with regular fetal heart rate. Rescue cervical cerclage by modified Shirodkar's technique was done under general anesthesia. She was administered prophylactic antibiotics and tocolytic. She was closely followed up at regular intervals and observed for signs of chorioamnionitis. She delivered at 35 weeks of gestation with two female children with birth weights 2.3 kg and 2.1 kg, respectively.

Keywords Cerclage, Cervical incompetency, Shirodker technique, Rescue cerclage

Background

Cervical cerclage is a surgical procedure that involves the placement of a suture around the cervix to prevent its premature opening. The procedure is typically performed between 12 and 16 weeks of pregnancy and can reduce the risk of pregnancy loss or preterm birth in women with cervical insufficiency. Rescue cervical cerclage is mostly performed in cases of threatened preterm delivery as a last resort to buy some time for fetal maturity and weight gain. Occasionally, it is done in early pregnancy with spontaneous cervical dilatation to salvage the pregnancy along with other efforts. During pregnancy, cervical cerclage is primarily done by techniques like McDonald's technique and Shirodkar's technique. There is one more technique called a modified Shirodkar's technique where the suture is placed as close as possible to the internal os by retracting the bladder from the anterior vaginal wall, but the posterior pouch is not opened.

Cervical cerclage has been a topic of extensive research in the past few decades, with several studies evaluating its efficacy, safety, and optimal timing. While the procedure is generally considered safe, there are potential risks and complications, such as infection, bleeding, rupture of the membranes, or cervical trauma. Therefore, it is crucial to identify the appropriate candidates for cervical cerclage and to weigh the benefits and risks carefully.

Additionally, there is ongoing debate about the optimal timing of the procedure, as some studies suggest that the effect of elective cerclage at 14–18 weeks of pregnancy is more ideal [1] and may provide better outcomes than cerclage done at a later stage of pregnancy. However, there is no consensus on the optimal timing, and the

*Correspondence:
Gaurav Sharma
drgauravsharma.bhandup@indiraivf.in
[1] Indira IVF Hospital Private Limited, Bhandup, Mumbai, India
[2] Samarth Maternity & Nursing Home, Mulund, Mumbai, India
[3] Indira IVF Hospital Private Limited, Dadar, Bhandup, Mumbai, India
[4] Indira IVF Hospital Private Limited, Udaipur, Rajasthan, India

decision should be individualized based on the patient's history, cervical length, and other risk factors. It is rare to perform a rescue cervical cerclage procedure as early as 8 weeks with exposed membranes, and so, this particular case makes it very important in saving a precious pregnancy.

This article aims to provide an overview of rescue cervical cerclage, including its indications, techniques, outcomes, and potential risks. By providing a comprehensive overview of cervical cerclage, this article aims to help clinicians make informed decisions about the management of cervical insufficiency and improve pregnancy outcomes for women with spontaneous cervical dilatation in the first trimester.

Case presentation

A 34-year-old primigravida conceived through IVF reported to our clinic with cramps and pain in the lower abdomen. She had conceived through IVF in our clinic through self-gametes and frozen embryo transfer. She did not have any history of surgery in the past or any significant medical history. Two grade 1 embryos (as per gardner's grading system) were transferred, and so she was carrying twins. By embryo transfer date, her pregnancy was of 8-week duration. Her previous TVS(Transvaginal sonography) was done 2 weeks earlier which revealed a twin pregnancy of 6 weeks, cervical length was 3.1 cm, and internal os closed without any signs of funnelling. The patient was alright until she started experiencing cramps in her lower abdomen previous night. She reported to the clinic in the morning complaining of cramps in her lower abdomen and blood-stained mucoid vaginal discharge.

On clinical examination, her vitals were stable, and she was afebrile. On per speculum examination, her cervix was dilated with amniotic membranes bulging into the cervical canal Blood-stained thick mucoid cervical discharge was noted (Fig.1b). On transabdominal USG, she

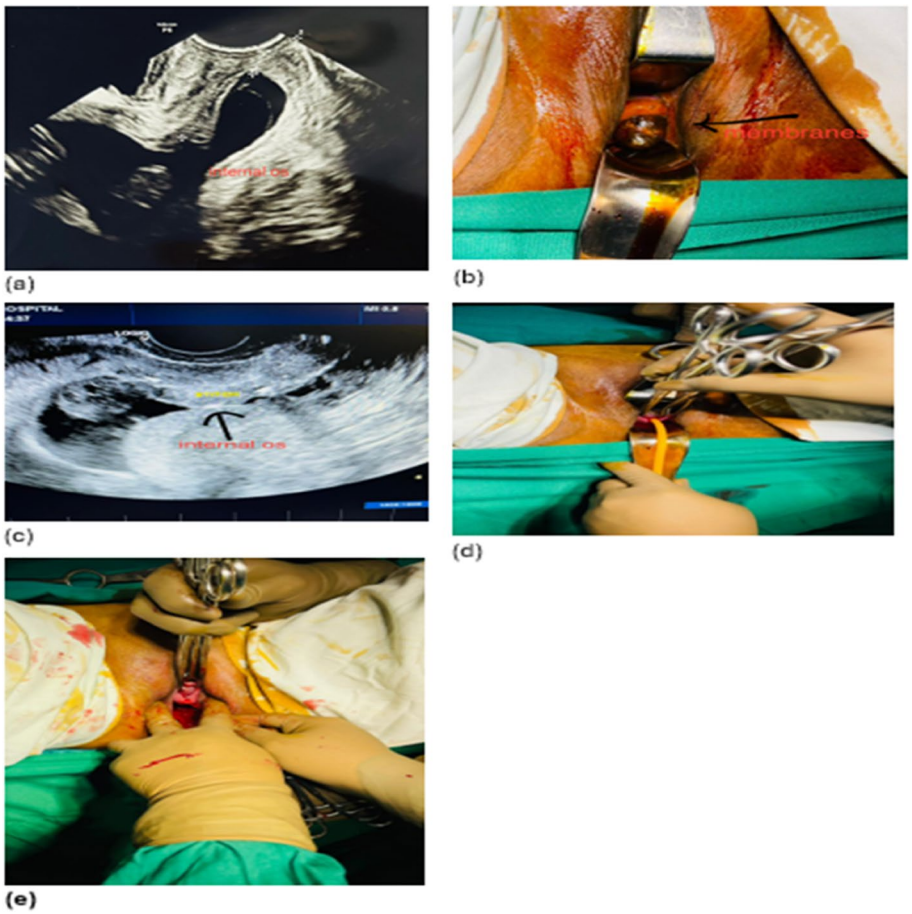

Fig. 1 **a** Preoperative TVS showing dilated internal os and membranes bulging into the vagina. **b** The per speculum pelvic examination shows membranes bulging through the internal os into the vagina. **c** The postoperative transvaginal sonogram showed a completely closed internal os after modifying Shirodkar's cerclage. **d** The Foley catheter with its bulb inflated was placed to prevent the membranes from bulging out during the procedure. **e** The image after the cervical cerclage knot was placed

had diamniotic dichorionic twin gestation. Both fetuses were of 8-week gestation. The fetal heart rate(FHR) of the 1st twin was 162 beats/minute, and the FHR of the second twin was 160 beats/minute. The gestational sacs were intact. The internal os was open, the internal os diameter was 1.5 cm as measured by USG, and membranes of the first twin were bulging out of the internal os.

The patient was hospitalized and counseled about the possibility of an inevitable abortion. She was administered inj. isoxsuprine 40-mg IV in 5% 500-mg dextrose at eight drops per minute as a tocolytic drug. She was counseled about the possible outcomes of the condition and the various treatment options. The rescue cerclage procedure was explained to her as a last resort, and she consented to the procedure. She was informed that the membranes could rupture any time before, during, or after the procedure, and the abortion could take place. She was also explained about the chances of chorioamnionitis and vertical transmission of infection. Vaginal swabs were taken to check for mycoplasma, chlamydia, and bacterial vaginitis.

Investigations

Her USG findings have been explained in the "Case presentation" and "Discussion" sections. Her preoperative hemoglobin was 12 g%, and leucocyte count was 12×10/L. Her CRP was 5 mg/L. Her postoperative blood test after 48 h of surgery revealed Hb 11.8 g%, WBC 9×10 to the power 9 please do need full. /L, and CRP was 7 mg/L. Her preoperative vaginal swab for mycoplasma, chlamydia, and bacterial vaginosis was negative. A repeat vaginal swab was taken after 7 days, and its reports were also negative for mycoplasma, chlamydia, and bacterial vaginitis. Her CRP after 7 days was 7 mg/L.

Treatment

The rescue cervical cerclage by modified Shirodkar's technique was performed under general anesthesia. It was a challenging procedure as the lip of the cervix could not be held with an instrument for fear of rupture of membranes since the membranes were bulging from the canal. After neatly exposing the cervix with Sims speculum, the membranes were gently pushed back through the cervical canal into the uterus using a sterile gauze. After that, the anterior end of the Foley catheter was placed in the cervical canal, and the bulb was inflated with 5 mL of normal saline so that membranes would not bulge out of the cervical canal again. Now, the anterior lip of the cervix was held with sponge-holding forceps, and hydro-dissection of the anterior vaginal wall was carried out. The bladder was pushed up, and a cervical stitch was placed at the level of the internal os with Marcelene tape. She was administered a single 1000-mg dose of amoxicillin intravenously. She was given 40 mg of isoxsuprine in 500-mg 5% dextrose at eight drops per minute during the first 24 h after the procedure. She did not complain of abdominal cramps after 24 h. The patient was kept under observation for the next 48 h and observed for any signs of infections like chorioamnionitis and leaking per viginum. She was prescribed an oral tab of isoxsuprine 10-mg TDS and oral antibiotics for 5 days. The patient was discharged after 48 h and asked to follow up after 5 days.

Follow-up and outcome

The patient followed up at weekly intervals for the next 4 weeks. The complete blood count and CPR were measured every week to see for the signs of infection and chorioamnionitis. Her leucocyte counts and CRP were within the normal range for her age and clinical condition. She was given weekly injections of depot hydroxyprogesterone intramuscularly until 4 weeks. The NT scan was done at 12 weeks, and NT was 2.9 mm in both twins. Her double marker tests revealed a low risk for trisomies 13, 18, and 21. She returned to routine activity after 4 weeks of surgery although with some restrictions. Further follow-ups were done at 4-week intervals until 28 weeks and at 2-week intervals until 34 weeks of gestation. There were no signs of fetal growth restriction. At 35 weeks, patient reported spontaneous premature rupture of membranes and underwent an emergency LSCS. The cervical stitch was removed in the same sitting. Two female children were delivered with baby weights of 2.3 kg and 2.1 kg, respectively. The babies were advised NICU admission by the pediatrician due to preterm delivery and breathing difficulty. The recovery in the NICU was uneventful. Their blood culture did not show any growth. They were in NICU for 7 days and then discharged when their clinical condition was stable. The babies were able to breastfeed on discharge.

It is imperative to note that this journey until live birth was possible only due to timely intervention and surgical intervention at 8 weeks of gestation and follow-up thereafter.

Discussion

The incidence of preterm spontaneous cervical dilatation is estimated to be about 1–2% of all pregnancies. However, it is difficult to determine the exact incidence because many cases go undiagnosed or misdiagnosed. The incidence was higher in women who had a history of preterm birth, a history of cervical surgery, or a history of cervical trauma [2]. According to a systematic review and meta-analysis published in the *Journal of Maternal–Fetal and Neonatal Medicine* in 2018, the incidence of cervical

incompetence in the first trimester was estimated to be approximately 0.1–0.5% of all pregnancies [3, 4].

ACOG recommends that cervical cerclage be considered for women with a history of spontaneous preterm birth, cervical length less than or equal to 25 mm on transvaginal ultrasound before 24 weeks of gestation, or a history of cervical surgery (level A recommendation) [5, 6]. However, there are no guidelines for rescue cervical cerclage.

Modified Shirodkar's cervical cerclage is a surgical technique to prevent premature delivery. It involves placing a nonabsorbable suture around the cervix to reinforce it and keep it closed. The procedure is typically performed between 12 and 16 weeks of gestation. It involves the dissection of the anterior vaginal wall to push the bladder above the internal os. The nonabsorbable suture like Mercelene tape is passed through the cervix at the level of internal os to give more strength to the internal os and prevent its dilatation. This technique is particularly difficult and requires a very skillful pelvic surgeon due to the risk of bladder injury, spontaneous labor, and excessive bleeding.

Rescue cervical cerclage has a very low success rate as compared to prophylactic cervical cerclage [5, 6]. The chances of rescue cervical cerclage being successful are very low in the first trimester. There are very few research articles on the success of rescue cervical cerclage in the first trimester. So, this successful case makes it very special for future reference. She was a primigravida, and there was no history of cervical surgery or injury. Her cervical length in the first scan at 6 weeks was 3.1 cm. It is important to note here that whenever a patient in the first trimester complains of cramping pain in the pelvic region, the suspicion of spontaneous cervical dilatation should be raised in a physician's mind. The most common cause of missed abortion or miscarriage in the first trimester is chromosomal disorders. But if cardiac activity is good, then uterine anatomical causes should be considered. In this case, if the membranes had ruptured, then it would have been impossible to save the pregnancy, but as long as the membranes are intact, the cervical cerclage can be attempted even if the membranes are bulging into the cervical canal.

Modified Shirodkar's technique of cervical cerclage has more chances of salvaging the pregnancy as the stitch is placed in a more physiological position as compared to McDonald's technique [7]. The cerclage procedure where membranes have been exposed requires a very high level of asepsis as the chances of chorioamnionitis following the procedure are very high. The patient should also be given tocolytic support to reduce uterine irritability and contractions. Antibiotic coverage before and after the procedure should be given to further reduce the chances of infection. Close follow-up of the patient should be maintained to check for early signs of sepsis or related infections. Regular monitoring of leucocyte counts and CRP levels should be done to pick up early signs of infection.

This case poses several key recommendations for obstetricians managing spontaneous cervical dilatation in the first trimester. Shirodkar's cervical cerclage is more physiological so it should be used in cervical cerclage. If the surgeon is not well trained, then modified Shirodkar's technique should be attempted. The obstetricians should take training in this technique. In recent years, McDonald's has gained more acceptance due to the short learning curve and fewer chances of surgical complications. However, it is pertinent to note that in McDonald's technique, it is not physiologically possible to take a knot at the level of internal os [8].

A second key recommendation is to check the diameter of internal os by USG when patients report to the clinic with cramping pain or vaginal discharge. If the internal os is found to be dilated, then a rescue cervical cerclage surgery can be recommended to the patient [6].

The third key recommendation is to check the internal os diameter [9] during routine obstetric scans. The TVS is much more effective in measuring internal os diameter as compared to the transabdominal route. Early detection of funnelling of internal os or premature dilatation can help in planning a rescue cerclage for the patient to prolong and salvage the pregnancy.

Learning points are as follows:

- This is a classic case of rescue cerclage with exposed membranes in the first trimester to salvage the pregnancy.
- Rescue cerclage should be attempted whenever there is premature cervical dilatation even if the membranes are bulging into the vagina.
- Shirodkar's cerclage is a very effective way of placing a cervical stitch at the level of internal os so it should be attempted. If the surgeon is not well trained in Shirodkar's technique, then modified Shirodkar's technique should be attempted as it is more physiological than McDonald's technique.
- Regular sonography check-ups of internal os diameter should be done during pregnancy to detect cases of funnelling of internal os or premature cervical dilatation.

Acknowledgements
We extend our heartfelt gratitude to Isha Suwalka for her invaluable assistance in proofreading the case report. Her meticulous attention to detail and expertise significantly enhanced the clarity and quality of our work. We are truly grateful for her dedication and support throughout the process.

Provenance and peer review
Not commissioned, externally peer reviewed.

Authors' contributions
GS contributed to writing most of the case report and detailed research regarding the "Discussion" section and the management strategy development. SRP was this case's primary surgeon and helped guide GS through the appropriate items to include in the case report. SZ was the main obstetrician involved in the patient's primary care until delivery. RS was involved in the treatment and decision-making of the patient's treatment. VC and NRB reviewed and revised the case report.

Funding
No funding received.

Availability of data and materials
The datasets used and/or analyzed during the current study will be available from the corresponding author upon reasonable request.

Declarations

Ethics approval and consent to participate
Indira IVF Hospital Private Limited Institutional Ethics Committee granted us a waiver stating that we do not need ethics approval (Reg. No. ECR/1629/Inst/MH/2021). The consent to participate is taken from the patient.

Consent for publication
The consent is taken from the participant for the publication.

Competing interests
The authors declare that they have no competing interests.

Received: 27 September 2023 Accepted: 23 March 2024
Published online: 30 April 2024

References

1. He D, Zhao D. Analysis of the timing of cervical cerclage treatment in pregnant women with cervical insufficiency and the effect on pregnancy outcome. Emerg Med Int. 2022;30(2022):8340009. https://doi.org/10.1155/2022/8340009. PMID: 35811604; PMCID: PMC9262559.
2. Voskamp BJ, Kok N, Schuit E, Kazemier BM, Oudijk MA. The incidence of cervical insufficiency in the Netherlands: a retrospective cohort study. Eur J Obstet Gynecol Reprod Biol. 2022;271:68–72. https://doi.org/10.1016/j.ejogrb.2021.11.010.
3. Alfirevic Z, Stampalija T, Roberts D, Jorgensen AL. Cervical stitch (cerclage) for preventing pregnancy loss in women. Cochrane Database Syst Rev. 2017;6:CD008991. https://doi.org/10.1002/14651858.CD008991.pub3.
4. Kollmann M, Gaulhofer J, Lang U, Klaritsch P, Herkner K, Klaritsch KU. Cervical insufficiency: a review of the literature. Geburtshilfe Frauenheilkd. 2021;81:601–7. https://doi.org/10.1055/a-1451-3529.
5. American College of Obstetricians and Gynecologists. Practice bulletin no. 231: cervical insufficiency. Obstet Gynecol. 2021;138(2):e49–61.
6. American College of Obstetricians and Gynecologists. Practice advisory: cervical insufficiency screening with cervical length measurement. Obstet Gynecol. 2020;136(4):e174–80.
7. Mohamed MA, El-Nemer AM, Hamid AM, et al. Shirodkar cervical cerclage: technique and outcomes. BMC Pregnancy Childbirth. 2017;17(1):93. https://doi.org/10.1186/s12884-017-1294-7.
8. Chiswick ML, Sinha P, Luxman D, et al. Shirodkar versus McDonald cerclage for cervical insufficiency: a systematic review and meta-analysis. BJOG. 2016;123(1):23–31. https://doi.org/10.1111/1471-0528.13580.
9. Yoon BH, Jun JK, Romero R, et al. Transvaginal sonographic assessment of cervical length and internal os diameter from midtrimester through the postpartum period in asymptomatic women with singleton fetuses. Am J Obstet Gynecol. 1997;176(3):703–10. https://doi.org/10.1016/s0002-9378(97)70626-9.

Publisher's Note