

CASE REPORT

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Late emergency cervical cerclage placement in a patient with early recurrent pregnancy loss and bicorporeal uterus: A case report

Asher Bashiri, Asaf Zaga, Amit Salinger Shpringer, Avishag Abecassis

ABSTRACT

Introduction: Uterine Müllerian anomalies are relatively common among the population, affecting approximately 4% of women, and can lead to various pregnancy-related pathologies. Cervical cerclage, a surgical procedure involving the placement of a stitch around the cervix, has been recognized as an optional treatment to mitigate the risk factors associated with these anomalies.

Case Report: A 33-year-old woman with a bicorporeal uterus and a history of early recurrent pregnancy loss, who was diagnosed with a cervical os shortening during her second anatomy screening at 22+3 weeks gestation. A cervical cerclage was performed to prevent preterm delivery, and subsequently. An elective cesarean section was conducted at 37+0 weeks of gestation, resulting in the birth of a healthy baby boy.

Conclusion: Although the literature about the effectiveness of cervical cerclage in pregnant women with Müllerian anomalies is limited, this case highlights its potential as an effective treatment option.

Keywords: Bicorporeal uterus, Cervical cerclage, Müllerian anomalies, Recurrent pregnancy loss

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INTRODUCTION

Uterine Müllerian anomalies are congenital disorders caused by abnormalities in development and fusion and/or absorption of the Müllerian ducts [1]. These anomalies are associated with several complications including ectopic pregnancies, preterm deliveries, premature rupture of membrane, cesarean delivery, breech presentation, recurrent pregnancy loss, and cervical os incompetence [1–3]. The anomalies occur in about 4% of the population, and among women with recurrent pregnancy losses, the prevalence varies between 15% and 42% [4, 5].

The direct cause of Müllerian anomalies is not yet completely understood, but there are several theories and hypotheses of polygenic and multifactorial causes, including mutation of the HOX10 gene [1].

Several classifications for Müllerian anomalies have been published during the last decade. The 2013 classification conducted by the European Society of Human Reproductive Embryology (ESHRE) divided the anomalies into six categories, while the 2021 classification published by the American Society for Reproductive Medicine (ASRM) classified them into nine different categories [6, 7].

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Researchers have explored the potential effectiveness of cervical cerclage as a preventive measure to reduce the increased risk of preterm delivery or late pregnancy losses associated with congenital uterine anomalies [8]. However, the efficacy of cervical cerclage in these cases remains unclear, except when there is a background of preterm delivery cases [5]. Cervical cerclage is a suture placed around the cervix, reinforcing its structure, and potentially reducing the risk of preterm delivery. Typically, there are several cerclage types including McDonald, Shirodkar, and modifications, and generally, the complications are usually rare; however, they include fistula formation and erosion of cerclage [9]. Due to limited research and conflicting evidence, the benefits and drawbacks of cervical cerclage in individuals with Müllerian anomalies must be evaluated according to specific cases.

In this case report, we present the complexity of treating a woman with Müllerian anomalies resulting in recurrent pregnancy loss, and we examine the role of cervical cerclage treatment for this patient to achieve live term delivery. This case report is clinically significant due to the controversies regarding cerclage in Müllerian anomalies. This case report is addressing the importance of discussing cerclage in woman with Müllerian anomalies early in pregnancy even without late pregnancy losses.

CASE REPORT

The patient was a 33-year-old female referred to our recurrent pregnancy losses prevention clinic at 5+0 weeks gestation following four previous early pregnancy losses, all of which occurred at the 8th week of gestation. The patient's medical history included a congenital hypoplastic right lung that caused asthma and right axis deviation of the heart. Despite her condition, she did not suffer from any symptoms or take any medications on a daily basis. Two of the patient's siblings were born with congenital hypoplastic right lungs as well.

Prior to her first visit in our clinic, the patient underwent a full recurrent pregnancy losses investigation (Table 1), which was unremarkable except for a bicorporeal uterus that was diagnosed after her first pregnancy loss with successful partial uterine septectomy.

Table 1: A panel of recurrent pregnancy losses investigation

Acquired thrombophilia
Lupus anticoagulant (LAC): 0.9 (0.8–1.20 ratio)
Anticardiolipin (ACL) antibodies IgM: 0.5 U/mL (0–20 U/mL)
Anticardiolipin (ACL) antibodies IgA: 0.6 U/mL (0–20 U/mL)
Anticardiolipin (ACL) antibodies IgG: Normal <1.6 U/mL (0–20 U/mL)
Antiβ2 glycoprotein 1 (Aβ2GP1) IgG: Normal <1.4 U/mL (0–20 U/mL)

Table 1: (Continued)

Antiβ2 glycoprotein 1 (Aβ2GP1) IgM: 0.6 U/mL (0–20 U/mL)
Hereditary thrombophilia
Factor V Leiden mutation: Normal
Prothrombin mutation: Normal
Antithrombin III: 88% (80–120%)
Protein S: 76% (60–150%)
Protein C: 79% (70–150%)
Hormonal and metabolic
TSH: 1.179 mU/L – (0.2–2.5 mU/L)
Anti-thyroglobulin: <20 IU/mL (10–40 IU/mL)
Anti-thyroid peroxidase: Normal >35 U/mL
Vitamin D levels: 56 nmol/L (recommended level 75–125 nmol/L)
Fasting glucose: 88 mg/dL (>95 mg/dL)
HbA1c: 5.1% (normal range <5.4%)
Immunologic
Rheumatoid factor: 15 IU/mL (0–30 IU/mL)
Anti-nuclear antibody (ANA): Negative
Genetic
Parental karyotypes: Normal (46 XX and 46 XY)
Anatomical investigation 3D sonography
Bicorporeal uterus class U3C (ESHRE classification 2013)

Her first physical examination in our clinic was normal. The patient's body mass index (BMI) was 25 (height 145 cm and weight 53 kg), and transvaginal sonography showed an intrauterine, single fetus with a heart rate; the crown-rump length (CRL) measurement was 10.1 mm, correlating to 7+1 weeks gestation. Our recommendations were to keep a healthy lifestyle, which included physical activity, stress reduction, and a decrease in caffeine consumption. In addition, supplementation of 1000 IU of Vitamin D, 400 mcg of folic acid, and 10 mg of dydrogesterone (Duphaston) twice per day until the 20th week of gestation was recommended.

Due to her congenital hypoplastic right lung, the patient consulted a pulmonologist who recommended that bronchodilators (fluticasone and vilanterol) be taken on a daily basis during her pregnancy.

At 13 weeks gestation, we discussed whether to place a cervical cerclage due to the patient's bicorporeal uterus. Eventually the decision was made not to place a cervical cerclage because the patient underwent a successful partial uterine septectomy and did not have any late pregnancy loss, preterm delivery, or cervical os incompetence in her obstetric history.

At 22+0 weeks gestation, the patient underwent a late anatomy scan that was normal; however, cervical length was barely measured at 3.4 mm (Figure 1). The patient was hospitalized immediately, and we discussed whether to place a late cervical cerclage due to concern

of a preterm delivery. We decided to place emergency McDonald cerclage, which was successfully preformed at 22+3 weeks gestation (Figure 2).

At 27 weeks gestation, the patient was diagnosed with gestational diabetes mellitus (GDMA1), which was well-controlled by diet. Due to the patient's preferences, obstetrics history, medical history, and habitus (height of 145 cm), a decision was made to perform an elective cesarean section with betamethasone (Celeston) treatment prior to the surgery.

At 37+0 weeks gestation, the patient underwent a successful elective cesarean section with the removal of the cerclage. A healthy baby boy was born with a normal Apgar score (9, 10 in 5 and 10 minutes) and a birth weight of 2315 g.



Figure 1: Transvaginal sonography of cervical length at 22+0 weeks gestation.



Figure 2: Transvaginal sonography of cervical length after cerclage placement.

DISCUSSION

The patient in the case had no history of late pregnancy loss or preterm delivery. The patient was diagnosed with bicorporeal uterus/U3 assessing the ESHRE classification [7] and classification bicornuate septate uterus by the ASRM classification [6]. Considering her obstetric history, the fact that she underwent partial

uterine septectomy and showed no evidence of cervical os incompetence or cervical shortening with a full panel of recurrent pregnancy loss (RPL) investigation that was unremarkable and led us to believe that we solved her main problem. Thus, we decided not to perform elective cervical cerclage. However, due to frequent monitoring and high-risk pregnancy follow-up provided by our RPL prevention clinic, a cervical insufficiency was diagnosed due to a measured cervical length of 3.4 mm during the patient's second anatomy screening at 22+0 weeks gestation. The diagnosis led to the performance of a late emergency cervical cerclage, eventually resulting in a successful term delivery at 37+0 weeks gestation for the patient.

It is still unknown at which percentiles Müllerian anomalies are associated with cervical os incompetence or cervical shortening, which is considered a leading cause of recurrent late pregnancy losses and preterm deliveries [2].

Airolidi et al. conducted a prospective study that revealed a significant 13-fold increase in the risk of preterm birth among patients with Müllerian anomalies and short cervix who underwent a series of cervical length examinations with ultrasound [10].

Although limited in number, several earlier studies, which comprised case-control studies and small cohorts and were conducted in the 1980s and 1990s, demonstrated the potential benefits of cervical cerclage in patients with Müllerian anomalies. In a study by Abramovici et al., 15 women with Müllerian anomalies who received cervical cerclage exhibited a remarkable 100% live birth rate, with only two cases of preterm deliveries [11]. Another study by Golan et al. found that approximately one-third of women with Müllerian anomalies experienced cervical os incompetence. The authors' research revealed that the placement of cervical cerclage significantly improved term deliveries and reduced rates of late pregnancy losses (26% to 63% and 74% to 37%, respectively) [12].

The impact of cervical cerclage is particularly significant among patients with a bicornuate uterus. Seidman et al. conducted a study that demonstrated a substantial increase in newborn survival rates among patients who underwent cervical cerclage (88% versus 47%). It is important to highlight that this study specifically excluded patients with arcuate and septate uteri, focusing solely on cases of bicornuate uterus [2]. Putting a cervical cerclage on a patient with bicorporeal uterus does not require specific technique since there is one cervix and the structure of bicorporeal uterus with septum is in the upper part of the uterus and not around the cervix. The placement of cerclage should be the same technique.

Leo et al. also suggested the preventive use of cervical cerclage for pregnancies with Müllerian anomalies; their results showed that term deliveries were five times higher after cervical cerclage placement [2]. Yassaee and Mostafee demonstrated the effectiveness of cervical cerclage in preventing preterm delivery in patients with

a bicornuate uterus. Their results showed a remarkable 76% term delivery rate in patients who underwent cervical cerclage compared to a mere 27% term delivery rate in patients without cerclage [5].

Timing of cervical cerclage as well as elective procedure versus emergency or rescue procedure is impotent for the successes. Several studies published recently [13, 14] have shown that elective cerclage has a better outcome, but late cerclage can also be considered. Thus, it is important to preform cerclage placement as an elective procedure if there is an indication and/or clinical finding.

Although the literature is minimal, many confounding variables exist that have the potential to cause bias. Possible factors include limited sample sizes, lack of definitional uniformity within the topic, differences in the timing of cerclage placement, and unclear screening criteria. These variables pose a challenge to making a definite determination about the necessity of cervical cerclage in patients with Müllerian anomalies [15].

This case report emphasizes that late placement of cerclage during pregnancy resulted in a successful delivery of a healthy baby. We advise to discuss the option of cerclage early in pregnancy, to consider its placement normally around 11–13 weeks gestation.

CONCLUSION

Considering this experience, we strongly recommend that our patient undergoes preventive cervical cerclage alongside high-risk pregnancy follow-up in her future pregnancies. Furthermore, we suggest discussing the option of cervical cerclage even after uterine septectomy with patients who have Müllerian anomalies, although the relevant literature is still controversial.

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Asher Bashiri – Conception of the work, Design of the work, Acquisition of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Asaf Zaga – Conception of the work, Design of the work, Acquisition of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Amit Salinger Shpringer – Design of the work, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Avishag Abecassis – Design of the work, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of

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Authors declare no conflict of interest.

Data Availability

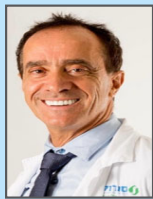
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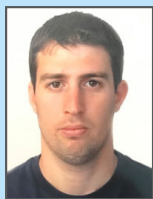
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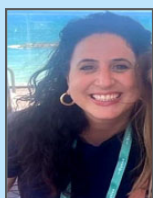
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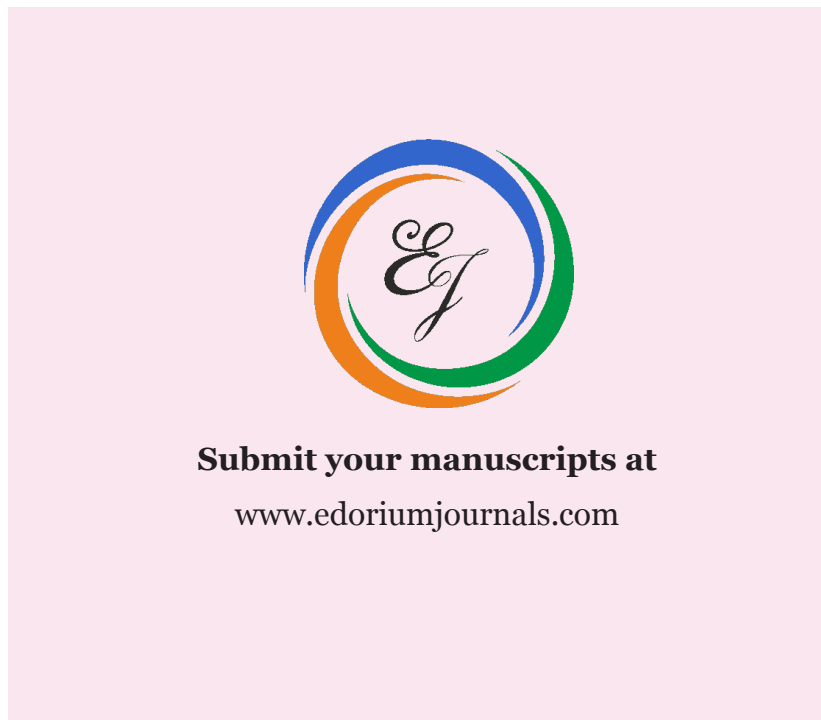
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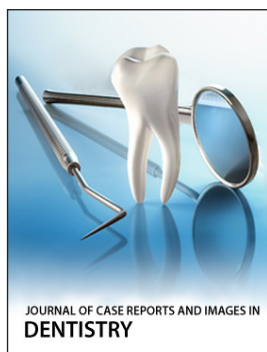
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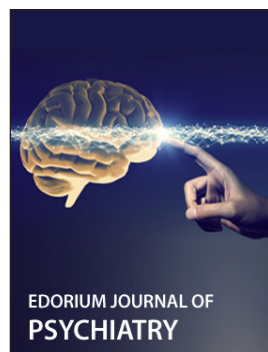
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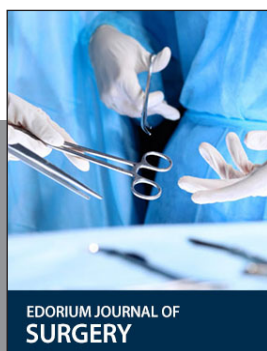
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