

# Effective treatment of cervical incompetence in a monochorionic monoamniotic twin pregnancy with a rescue cervical cerclage and pessary – a case report and review of literature

Niewydolność szyjki macicy u pacjentki w ciąży bliźniaczej jednokosmówkowej jednoowodniowej skutecznie leczona szwem szyjkowym ratunkowym i pessarium – opis przypadku i przegląd piśmiennictwa

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## Abstract

*A monochorionic monoamniotic (MCMA) twin pregnancy is the rarest form of twin gestation, accounting for around 1:10000 to 1:20000 of all deliveries regardless of the region of the world. All multiple gestations have a higher risk of preterm delivery due to either preterm uterine contractions or asymptomatic cervical shortening (cervical incompetence).*

*A case of a 28-year-old primigravida in MCMA twin pregnancy with cervical incompetence diagnosed at 22 weeks of gestation is presented. After obtaining cervical swabs, negative laboratory infection parameters and confirming concordant gestational age on ultrasound scan with no structural abnormalities of both fetuses, the patient was qualified for an emergency cervical cerclage according to Wurm-Hefner method. Five days after the procedure, a cervical pessary was additionally inserted. She was administered antibiotics and steroids. The wellbeing of both fetuses was strictly monitored by means of cardiotocography tracing and ultrasound examinations, on which they were both eutrophic, with no abnormalities in Doppler blood flow patterns. Spontaneous premature rupture of membranes took place at 32 weeks of gestation, a cesarean section was performed and two female fetuses of 1740g and 1760g were delivered. They both required antibiotics because of congenital pneumonia, but no respiratory support was necessary. The twins were discharged from the hospital 22 days after birth in good general condition. This case of a rescue cervical cerclage and pessary used simultaneously can be an example of an effective method of cervical incompetence treatment in twin pregnancies.*

Key words: **monochorionic monoamniotic twin pregnancy / uterine cervical incompetence / emergency / rescue cervical cerclage / cervical pessary / preterm delivery /**

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## Streszczenie

Ciąża bliźniacza jednokosmówkowa jednoowodniowa (JKJO) jest najrzadszym wariantem ciąży bliźniaczej. Porody ciąż jednoowodniowych stanowią około 1:10000 do 1:20000 wszystkich porodów niezależnie od regionu świata. Ciąże mnogie obarczone są wyższym ryzykiem porodu przedwczesnego w mechanizmie przedwczesnej czynności skurczowej macicy lub bezobjawowego skracania i niewydolności cieśniowo-szyjkowej.

Przedstawiono przypadek 28-letniej pierwiastki w ciąży bliźniaczej JKJO, u której zdiagnozowano niewydolność cieśniowo-szyjkową w 22. tygodniu ciąży. Po pobraniu posiewów z kanału szyjki macicy oraz uzyskaniu negatywnych wykładników stanu zapalnego, pacjentkę zakwalifikowano do założenia ratunkowego szwu szyjkowego metodą Wurm-Hefnera.

Pięć dni po operacji dodatkowo założono pessar szyjkowy. Zastosowano antybiotykoterapię oraz sterydoterapię. Dobrostan obu płodów ściśle monitorowano przy użyciu kardiotokografii oraz badań ultrasonograficznych, w których potwierdzono prawidłowy rozwój płodów oraz brak odchyłań w badaniach dopplerowskich. W 32. tygodniu ciąży samoistnie odplynął płyn owodniowy. Pacjentkę rozwiązano cięciem cesarskim.

Urodzono dwa noworodki płci żeńskiej o masie 1740g oraz 1760g, które wymagały antybiotykoterapii z powodu wrodzonego zapalenia płuc. Niemniej jednak nie wymagały żadnego wsparcia oddechowego. Bliźnięta wypisano do domu 22 dni po porodzie w stanie ogólnym dobrym. Prezentowany przypadek zastosowania szwu szyjkowego ratunkowego oraz pessarium może stanowić przykład skutecznej metody leczenia niewydolności cieśniowo-szyjkowej w ciąży bliźniaczej.

Słowa kluczowe: **ciąża bliźniacza, jednokosmówkowa, jednoowodniowa /  
/ ratunkowy szew szyjkowy / pessar szyjkowy / poród przedwczesny /**

## Introduction

A monochorionic monoamniotic (MCMA) twin pregnancy is the rarest form of twin gestation, accounting for 1% of monozygotic pregnancies. The rising trend of multiple gestations in the world pertains mostly to dizygotic pregnancies and is related to the age of the mother, race or the use of different methods of assisted conception, whereas the percentage of monozygotic pregnancies remains stable. MCMA deliveries constitute around 1:10000 to 1:20000 of all deliveries, regardless of the region of the world [1, 2].

MCMA gestation is a high risk pregnancy due to a variety of complications. The percentage of fetal and neonatal demise may be as high as 60%. The above is mainly due to prematurity, fetal anomalies and pathologies typical for monochorionic pregnancies [1]. Umbilical entanglement is a complication specific only for MCMA pregnancy. Its frequency is estimated to be around 80% [3, 4]. As a result, the world and Polish Gynecological Societies recommend an elective cesarean section at 32 weeks of gestation, after applying steroid therapy [5, 6].

Regardless of chorionicity, all multiple gestations have a higher risk of preterm delivery due to either preterm uterine contractions or asymptomatic cervical shortening (cervical incompetence). According to Chauhan, the risk of preterm delivery in multiple gestation is six-fold higher in comparison to singletons and of low birth weight (LBW <2500g) and very low birth weight (VLBW <1500g) neonate is eight times higher [7]. The article presents a case report of a MCMA pregnancy, complicated by cervical insufficiency, successfully treated with a rescue cervical cerclage and pessary.

## Case report

A 28-year-old patient was admitted to the 1<sup>st</sup> Department of Obstetrics and Gynecology, Medical University of Warsaw, at 22 weeks of her first MCMA twin gestation, with the diagnosis of cervical incompetence.

The pregnancy resulted from a natural conception. The history of uterine cervix surgical procedures was negative. The pregnancy was uneventful until 20 weeks. The cervical length assessed at 18 weeks of gestation was 28mm. During a routine outpatient visit at 22 weeks, a dilation of 3 cm was found and the inferior pole of the amniotic sac was protruding into the vagina. Following admission, laboratory infection parameters were assessed, a cervical swab was taken and a test for IGFBP1 (Insulin like growth factor binding protein 1) in the cervical discharge was performed. Laboratory results showed white blood cells count (WBC) 16.4 G/l and C-reactive protein (CRP) 2.6 mg/l. The cervical culture was positive for *Escherichia coli*, *Enterococcus faecalis* and *coagulase-negative Staphylococcus* and the presence of IGFBP1 was confirmed. The biometry of both fetuses was concordant with gestational age and there were no structural abnormalities on the ultrasound scan. The amniotic fluid index was normal and Doppler blood flow pattern in both umbilical arteries, middle cerebral arteries and ductus venosus were within the normal ranges.

The patient was informed about the prognosis for the pregnancy. After obtaining an informed consent she was qualified for a rescue cervical cerclage. On the first day of hospitalization, at 22 weeks of gestation, a Wurm-Hefner cerclage was placed in Trendelenburg position, under general anesthesia, after pushing up the amniotic sac by filling the urinary bladder with 0.9% sodium chloride (NaCl) solution. There were no intra- and early postoperative complications. Tocolytic therapy with intravenous infusion of fenoterol was administered for 24 hours in the postoperative period. In the perioperative period prophylactic antibiotic therapy with ampicillin/sulbactam was administered, followed by metronidazole with erythromycin for 10 days. The perioperative period was uneventful. Five days after the procedure, a cervical pessary was additionally inserted in order to decrease the pressure of amniotic sac on the internal cervical os. Bed rest was introduced - the patient was allowed to stand up only

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for meals and toilet/shower. On the 19<sup>th</sup> day after the procedure partial slipping down of the cerclage was diagnosed and amniotic sac was again visible protruding into the vagina. Steroids were administered twice – at 24 and 29 weeks of gestation. Laboratory tests showed no signs of infection. Nevertheless, *E.coli* was cultured in repeated cervical swabs. The wellbeing of both fetuses was strictly monitored by means of cardiotocography tracing and ultrasound examinations, on which they were both eutrophic, with no abnormalities in Doppler blood flow patterns. However, a probable site of umbilical cords collision with normal Doppler flow in umbilical arteries and veins was observed. (Figure 1, Figure 2).

There was no progress in cervical dilation during the next six weeks. Spontaneous premature rupture of membranes took place at 32 weeks of gestation. The patient was administered 4g of intravenous magnesium sulphate and cesarean section was performed. Two female fetuses of 1740g and 1760g were delivered in good general condition and a double true umbilical knot was found on both umbilical cords. The operation and postoperative period were uneventful. Congenital pneumonia was diagnosed in both neonates (probably due to premature rupture of membranes) and they were admitted to the neonatal intensive care unit (NICU). Antibiotics were administered, but no respiratory support was required. The twins were discharged from NICU after 11 days and discharged home together with the mother 22 days after birth in good general condition.

## Discussion

Preterm delivery, defined as a delivery before 37 completed weeks of gestation, affects half of the twin pregnancies - 53% of twin pregnancies are delivered before 37 weeks, 16% before 34 weeks and 4% before 28 weeks of gestation [8]. The etiology of preterm shortening and dilation of the cervix has been studied for many years. It could be the result of surgical procedures performed on the cervix, congenital uterine and cervical malformations, effect of diethylstilbestrol during fetal life or an ongoing inflammatory process [9]. The latter has been under thorough investigation. Patients at high risk of preterm delivery have increased levels of interleukins: IL-1, IL-6, IL-8, IL-10; TNF (tumor necrosis factor) and glucose in the amniotic fluid, as well as fetal fibronectin and IGFBP1 in the cervical discharge. However, in twin pregnancies, even if the fetal fibronectin test is negative and the cervical length is >20mm at 20 weeks of gestation, 10.3% of patients will still deliver before 34 weeks [7]. Even though screening tests are applied, there is a group of patients with no additional risk factors and asymptomatic dilation of the cervix diagnosed at a stage, when the amniotic sac is already protruding into the vagina. Cervical cerclage is one of the treatment options of cervical insufficiency in a singleton pregnancy [10]. It is an effective procedure if performed according to specific indications. However, cervical cerclage is not recommended for twin pregnancies. It has been proven that in multiple pregnancies it does not reduce the risk of preterm delivery [11, 12], in fact it may even increase it (RR 2.15) [13].

In case of cervical dilation and amniotic sac protrusion, an emergency cervical cerclage should be taken into consideration if vaginal bleeding, amniotic fluid leakage, symptoms of chorioamnionitis, the risk of hypoxia and fetal anomalies were excluded. Published data on rescue cervical cerclage in



Figure 1. Ultrasound scan of cord entanglement.

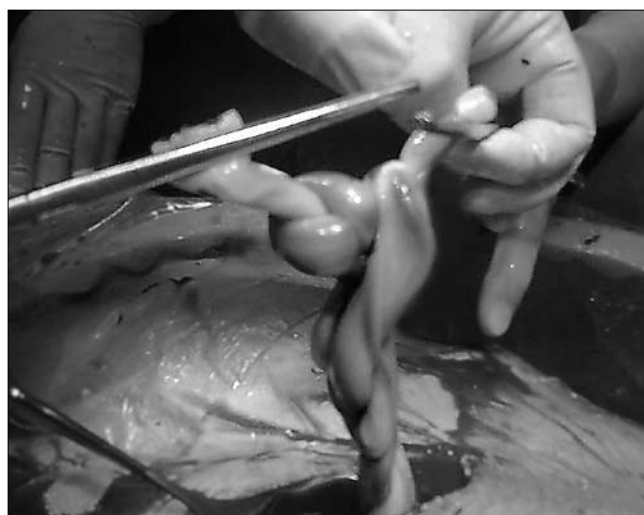


Figure 2. Cord entanglement.

twin pregnancies are scarce. As it does not affect the neonatal mortality and morbidity rates and might cause complications, the qualification for the procedure has to be individual in each case and an informed consent ought to be signed by the patient.

Cervical cerclage procedure was performed for the first time in the case of cervical incompetence by Shirodkar in 1955 in Bombay [14]. Since then, many researchers have tried to assess the effectiveness of different cervical cerclage techniques in singleton pregnancies. Indications and contraindications were established to identify a group of patients who would benefit most from such a procedure. The efficacy of an emergency cervical cerclage was also evaluated in clinical studies. Cerclage significantly prolongs the duration of pregnancy in comparison to bed rest. However, it does not reduce neonatal mortality and morbidity rates [15, 16].

Moreover, there are no published randomized studies assessing the effectiveness of rescue cervical cerclage in multiple gestations. Mahendira presented the results of 28 rescue cervical

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cerclages in twins and triplets. In case of twins, the average time from the procedure to delivery was 9 weeks. Patients usually delivered around 31 weeks of gestation. There were no neonatal deaths, 82% of the newborns required admission to the neonatal intensive care unit due to prematurity [17]. Overton and Warren described a triplet pregnancy with intrauterine demise of one fetus at 15 weeks of gestation, where the emergency cerclage procedure allowed the delivery at 29 weeks [18]. Khan described a delivery of a second fetus at term after the miscarriage of the first twin at 21 weeks of gestation and the use of a rescue cervical cerclage [19]. There is also a case of a multiple pregnancy complicated by cervical incompetence in Polish literature. The rescue cervical cerclage was used at 22 weeks of gestation and the delivery took place at 34 weeks [20]. In our patient the time from the procedure to delivery was 10 weeks, which is consistent with the results presented by Mahendira. However, a pessary was also placed and, to the best of our knowledge, the combination of the two methods has not yet been described in the literature.

There are many different techniques of the cervical cerclage procedure. Wurm-Hefner method was used in our patient. However, emergency cerclages may also often be performed according to the McDonald method [21, 22]. In order to achieve the retraction of the amniotic sac from the external cervical os, our patient was laid in the Trendelenburg position and the urinary bladder was filled with NaCl solution. Different methods of amniotic sac retraction are described in the literature. Ultrasound-guided amniocentesis may be used to reduce the pressure applied by the sac [23]. A placement of a transcervical balloon in the lower part of the uterus to push back the amniotic sac was also described [24]. Nevertheless, there have been no studies comparing the two methods so far.

While assessing the effectiveness of the emergency cerclage in singleton pregnancies, the prognostic factors were established. Cervical dilation of more than 4 cm is related to a high risk of failure. C reactive protein >4mg/l and WBC >14 000 G/l are related to an increased risk of miscarriage/delivery, despite the use of a cerclage [25, 26]. However, there are no similar reports concerning multiple pregnancies. Our patient had a 3-cm dilation, WBC was increased but CRP was 2.6mg/l. The pathogens isolated from the cervix might have indicated an infection that could have been one of the etiological factors of cervical incompetence. In case of intrauterine infection and chorioamnionitis, the risk of preterm delivery and fetal infection increases. Higher concentrations of IL-6, metalloproteinase 8 (MMP8) and TNF $\alpha$  are the risk factors contributing to ineffectiveness of cerclage [27, 28]. The level of IL-6 strongly correlates with the procedure-delivery interval [29].

In the presented case intensive antibiotic therapy was applied. Even though *E. coli* was isolated in consecutive cultures from cervical swabs, parameters of inflammation did not increase and there was no further dilatation of the cervix. Nevertheless, spontaneous premature rupture of membranes at 32 weeks of gestation indicated that there had been an ongoing infection.

## Conclusion

This case of a rescue cervical cerclage and pessary used simultaneously can be an example of an effective method of cervical incompetence treatment in twin pregnancies.

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