## Cervical insufficiency in twin pregnancy – case report of a successful physical examination-indicated cerclage

# Incompetência cérvico-ístmica em gravidez gemelar – caso de sucesso de ciclorrafia com indicação pelo exame físico

Ana Isabel Cunha¹, Filipa Brás², Elsa Pereira³, José Manuel Furtado⁴ Hospital Senhora da Oliveira – Guimarães

### **Abstract**

Preterm birth rate is increased in twin pregnancies and there is currently no proven effective intervention for the prevention of this complication. The authors present a case of cervical insufficiency in a twin gestation identified in the  $21^{st}$  week. Although the patient was asymptomatic, the cervix was dilated with exposed membranes and physical examination-indicated cerclage was performed. The cervical length remained stable over the rest of the pregnancy. Gestation was terminated in the  $35^{th}$  week due to fetal growth restriction in fetus 2. Both fetuses had an Apgar score of 9/10/10 at 1,5 and 10 minutes post-birth.

**Keywords:** Twin pregnancy; Cervical insufficiency; Cerclage.

#### **INTRODUCTION**

C ervical insufficiency is the incapacity of the uterine ne cervix to sustain a pregnancy in the second trimester in the absence of contractions<sup>1</sup>. Its pathophysiology is still poorly understood. Although structural cervical weakness may be the cause of many recurrent second-trimester abortions, the majority of cases are probably caused by other disorders such as decidual infection, placental bleeding or uterine overdistension, as is usually the case in multiple pregnancies<sup>2</sup>.

In Portugal, in 2018, multiple births accounted for 1,6 percent of total births with 62,6 percent of these births being preterm<sup>3</sup>. Twin pregnancy is associated with higher rates of several potential complications of

Several interventions, including vaginal progesterone, pessary or placement of a cervical cerclage, have been proposed to reduce the risk of preterm birth in twin gestations but, to date, none has clearly proven its effectiveness<sup>2,4</sup>. In a 2017 meta-analysis by Jarde et al.<sup>4</sup> that assessed the effectiveness of cerclage or pessary for preventing preterm birth the authors concluded that no intervention was beneficial for this purpose. Despite the routine use of these procedures in the management of twin pregnancies may be ineffective, the judicious utilization of some interventions in selected clinical circumstances may be of inestimable value, such as physical examination-indicated cerclage placement.

The authors describe a case of cervical insufficiency in a twin pregnancy identified in the 21<sup>st</sup> week of gestation in which the performance of a physical examination-indicated cerclage resulted in good maternal-fetal outcomes. For the elaboration of this article, an informed consent was obtained from the patient.

pregnancy of which the most serious is preterm delivery. Besides neonatal death, complications of prematurity include respiratory distress syndrome, intraventricular hemorrhage, necrotizing enterocolitis, retinopathy of prematurity, among others.

<sup>1.</sup> Interna de Formação Específica de Ginecologia e Obstetrícia, Hospital da Senhora da Oliveira – Guimarães.

<sup>2.</sup> Assistente Hospitalar de Ginecologia e Obstetrícia, Hospital da Senhora da Oliveira – Guimarães

<sup>3.</sup> Assistente Hospitalar Graduada de Ginecologia e Obstetrícia, Hospital da Senhora da Oliveira – Guimarães.

<sup>4.</sup> Diretor de Serviço de Ginecologia e Obstetrícia, Hospital da Senhora da Oliveira – Guimarães.

#### **CASE REPORT**

A 25-year-old patient, primigravida with no relevant medical/gynecological history, was referred for surveil-lance due to a spontaneous dichorionic twin pregnancy. In the first trimester ultrasound, no structural anomalies were detected and the screening for fetal chromosomal abnormalities was negative for both fetuses.

In the 17<sup>th</sup> week, although the patient was asymptomatic, the cervical length was 25 mm. We decided to initiate vaginal progesterone to reduce the risk of preterm birth. There were no signs of urinary tract infection as demonstrated by negative urine culture.

The  $2^{nd}$  trimester ultrasound, at 21 weeks and 2 days of gestation, showed normal morphology in both fetuses and amniotic fluid superior to normal, yet not reaching the threshold for diagnosing polyhydramnios. The transvaginal ultrasound (Figure 1) demonstrated a cervical length of 6 mm with pronounced funneling and in the speculum examination the cervix was dilated with exposed membranes. The patient maintained no symptoms of clinical contractions.

After discussion with the couple about the prognosis and potential interventions to decrease the risk of preterm labor, the decision was to perform a cervical cerclage. The procedure was executed at 21 weeks and 5 days of pregnancy using McDonald's technique (Figure 2). Indomethacin 25 mg every 6 hours was prescribed for 24 hours post-procedure.

In the  $22^{nd}$  week, although the pregnant woman remained asymptomatic, the speculum examination raised suspicion of the presence of amniotic fluid in the posterior pouch of vagina. There was no leakage of amniotic fluid through the external cervical os using the Valsalva maneuver. The cervical length was 4-6 mm and the suture was visible on the distal third of its extent. The patient was treated with ampicillin. During hospitalization, she remained asymptomatic and there were not any other signs of amniotic fluid loss. The patient was discharged after 7 days of surveillance.

On the  $3^{rd}$  trimester ultrasound, performed in the  $28^{th}$  week, the cervical length evaluation (Figure 3) was similar to previous measurements (4-6 mm) and there were no clinical signs of uterine contractions.

In an ultrasound performed at 35 weeks and 5 days, the fetus 1 had an estimated fetal weight in the percentile 4 and normal Doppler evaluation and the fetus 2 had an estimated fetal weight in the percentile 0,5 with altered cerebroplacental ratio. We decided to terminate pregnancy and a cesarean section was perfor-



**FIGURE 1.** Transvaginal ultrasound at 21 weeks and 2 days: cervix with a length of 6 mm and pronounced funneling.



**FIGURE 2.** Cervical length of 4-6 mm at 21 weeks and 5 days (transvaginal ultrasound before the placement of cerclage).



**FIGURE 3.** Transvaginal ultrasound in the 28th week showing cerclage suture in the cervix and a cervical length of 4-6 mm.

med due to breech presentation of fetus 2. Fetus 1 weighted 2310 grams at birth and fetus 2 weighted 1810 grams. Both had an Apgar score of 9/10/10 (at 1, 5 and 10 minutes post-birth respectively). After the procedure the cerclage was removed.

### **DISCUSSION**

The authors report a case of successful prenatal management with cerclage placement of a twin pregnancy that presented with cervical dilation in the  $21^{\rm st}$  week.

In the present case, the first warning sign was a short cervix detected in the  $17^{th}$  week. In our practice we screen for short cervical length in multiple gestation since it is a good predictor of preterm birth in these pregnancies<sup>5,6</sup> and initiate supplementation with vaginal progesterone when the cervix is short ( $\leq 25$  mm). In a 2017 meta-analysis from six randomized trials of asymptomatic women with twin gestations and a cervical length  $\leq 25$  mm, vaginal progesterone reduced the risk of preterm birth <33 weeks of gestation by 31% and neonatal death by  $47\%^7$ . The relative risks of neonatal death, respiratory distress syndrome and birth weight <1500g were also reduced significantly, without any demonstrable deleterious effects on childhood neurodevelopment.

In the 21st week, the patient presented with cervical dilation in speculum examination. Cervical dilation in the mid-trimester is associated with a more than 90% rate of spontaneous preterm birth and a poorer perinatal prognosis<sup>8,9</sup>. Although there are potentially beneficial interventions for prevention of preterm birth, the treatment options in the presence of cervical dilation are limited to physical examination-indicated cerclage (also known as emergency or rescue cerclage). After informing the couple about the unfavorable prognosis and discussing the available evidence about managing options, the decision was to place a rescue cervical cerclage.

The only randomized controlled trial to date that determined if physical exam indicated cerclage reduces the incidence of preterm birth in women with twin gestations and cervical dilation (1-5 cm) found a significant decrease in preterm birth at all evaluated gestational ages (including a 50% decrease in preterm birth <28 weeks) and a 78% decrease in perinatal mortality<sup>10</sup>. In this study, all women who underwent cerclage received prophylactic antibiotics and indomethacin. This was a small sample size study that was terminated early because of the significant decrease in perinatal mortality in the cerclage group.

In a 2019 meta-analysis by Li et al.<sup>11</sup>, which included 1211 women, the authors concluded that cerclage placement is beneficial for the reduction of preterm birth and the prolongation of pregnancy in twin pregnancies with a cervical length <15 mm or dilated cer-

vix >10 mm. The benefit was not demonstrated for history-indicated or twin alone-indicated cerclage. In women with cervical dilation, cerclage placement was associated with a significant prolongation of pregnancy, reduction of preterm birth rate and improvement in perinatal outcomes.

Earlier meta-analyses showed no benefit of cerclage for women with twin pregnancies<sup>2,4,12</sup>. A 2014 Cochrane review4 that included five randomized controlled trials (two of obstetric history indicated-cerclage and three of cerclage indicated by transvaginal ultrasound) reported no differences in perinatal deaths, serious neonatal morbidity or preterm birth rates when cerclage was compared with no cerclage in women with twin gestations. Saccone et al.12 evaluated the efficacy of cerclage in twin pregnancies with a short cervical length and found no differences in preterm birth <34 weeks and perinatal deaths. Very low birthweight and respiratory distress syndrome were more frequent in the cerclage group. Since these meta-analyses included few and small clinical studies, their conclusions are of limited value and need to be interpreted with caution.

Recently, several studies demonstrated a potential benefit of physical examination-indicated cerclage in reducing preterm birth rate in twin gestations<sup>13-16</sup>. Roman et al.13 reported that an association of cerclage, indomethacin and antibiotics in twin pregnancies with cervical dilation ≥ 1 cm was associated with a significant prolongation of gestation, decreased incidence of preterm birth and improved perinatal outcome, compared with expectant management. In a 2018 retrospective cohort study of twin pregnancies14, cerclage placement significantly reduced the incidence of preterm birth <32 weeks compared with expectant management only in the subgroup of rescue cerclage. Abbasi et al. 15 compared the perinatal outcomes in twin pregnancies with cervical dilation treated with cerclage or expectant management and observed lower rates of preterm birth, neonatal intensive care unit admission and neonatal respiratory morbidity in the cerclage group. Importantly, in this study, all women managed expectantly gave birth prior to 28 weeks.

Other authors compared the outcomes of emergency cerclage in twin versus singleton gestations and concluded that the beneficial obstetric outcomes are similar between the two groups<sup>17-20</sup>.

In conclusion, preterm birth is one of the main challenges in the management of multiple pregnancies. Whilst it is the major source of perinatal morbidity and mortality in twin gestations, there is currently no

proven effective intervention for the prevention of this complication. Despite the need for more high-quality trials of appropriate size and duration, physical examination-indicated cerclage may be an option for preventing preterm birth in twin pregnancies.

#### REFERENCES

- 1. American College of Obstetricians and Gynecologists. ACOG Practice Bulletin No.142: Cerclage for the management of cervical insufficiency. Obstet Gynecol 2014; 123(2 Pt 1): 372-379.
- 2. Rafael TJ, Berghella V, Alfirevic Z. Cervical stitch (cerclage) for preventing preterm birth in multiple pregnancy. Cochrane Database Syst Rev 2014;9:CD009166.
- 3. Instituto Nacional de Estatística- Estatísticas da Saúde: 2018. Lisboa: INE, 2020.
- 4. Jarde A, Lutsiv O, Park CK, et al. Preterm birth prevention in twin pregnancies with progesterone, pessary, or cerclage: a systematic review and meta-analysis. BJOG 2017; 124: 1163–1173.
- 5. Conde-Agudelo A, Romero R, Hassan SS, Yeo L. Transvaginal sonographic cervical length for the prediction of spontaneous preterm birth in twin pregnancies: a systematic review and metaanalysis. Am J Obstet Gynecol 2010; 203(2): 128.e1–128.12.
- 6. Lim AC, Hegeman MA, Huis In 'T Veld MA, et al. Cervical length measurement for the prediction of preterm birth in multiple pregnancies: a systematic review and bivariate meta-analysis. Ultrasound Obstet Gynecol 2011; 38:10–17.
- 7. Romero R, Conde-Agudelo A, El-Refaie W, et al. Vaginal progesterone decreases preterm birth and neonatal morbidity and mortality in women with a twin gestation and a short cervix: an updated meta-analysis of individual patient data. Ultrasound Obstet Gynecol 2017; 49: 303–314.
- 8. Daskalakis G, Papantoniou N, Mesogitis S, Antsaklis A. Management of cervical insufficiency and bulging fetal membranes. Obstet Gynecol 2006; 107(2 Pt 1): 221-226.
- 9. Althuisius SM, Dekker GA, Hummel P, van Geijn HP. Cervical incompetence prevention randomized cerclage trial: emergency cerclage with bed rest versus bed rest alone. Am J Obstet Gynecol 2003; 189(4): 907-910.
- $10.\ Roman\ A,$  Zork N, Haeri S, et al. Physical examination-indicated cerclage in twin pregnancy: a randomized controlled trial. Am J Obstet Gynecol 2020.

- 11. Li C, Shen J, Hua K. Cerclage for women with twin pregnancies: a systematic review and metaanalysis. Am J Obstet Gynecol 2019; 220: 543-557.
- 12. Saccone G, Rust O, Althuisius S, Roman A, Berghella V. Cerclage for short cervix in twin pregnancies: systematic review and metaanalysis of randomized trials using individual patient-level data. Acta Obstet Gynecol Scand 2015; 94: 352-358.
- 13. Roman A, Rochelson B, Martinelli P, et al. Cerclage in twin pregnancy with dilated cervix between 16 to 24 weeks of gestation: retrospective cohort study. Am J Obstet Gynecol 2016; 215: 98. e1-11.
- 14. Han MN, O'Donnell BE, Maykin MM, Gonzalez JM, Tabsh K, Gaw SL. The impact of cerclage in twin pregnancies on preterm birth rate before 32 weeks. J Matern Fetal Neonatal Med 2018: 1-9.
- $15.\,Abbasi$  N, Barrett J, Melamed N. Outcomes following rescue cerclage in twin pregnancies. J Matern Fetal Neonatal Med 2018;31: 2195–2201.
- 16. Pan M, Zhang J, Zhan, et al. Physical examination-indicated cerclage in twin pregnancy: a retrospective cohort study. Arch Gynecol Obstet 2020; Epub ahead of print.
- 17. Bernabeu A, Goya M, Martra M, et al. Physical examination-indicated cerclage in singleton and twin pregnancies: maternal-fetal outcomes. J Matern Fetal Neonatal Med 2016; 29: 2109-2113.
- 18. Miller ES, Rajan PV, Grobman WA. Outcomes after physical examination-indicated cerclage in twin gestations. Am J Obstet Gynecol 2014; 211: 46.e1–5.
- 19. Park JY, Cho SH, Jeon SJ, et al. Outcomes of physical examination-indicated cerclage in twin pregnancies with acute cervical insufficiency compared to singleton pregnancies. J Perinat Med 2018; 46: 845–852.
- 20. Rebarber A, Bender S, Silverstein M, et al. Outcomes of emergency or physical examination-indicated cerclage in twin pregnancies compared to singleton pregnancies. Eur J Obstet Gynecol Reprod Biol 2014; 173: 43–47.

#### **ENDEREÇO PARA CORRESPONDÊNCIA**

Ana Isabel Cunha

E-mail: anaisabelpcunha@gmail.com

**RECEBIDO EM:** 27/03/2021

**ACEITE PARA PUBLICAÇÃO:** 16/09/2021