

Angular pregnancy – an unexpected diagnosis

Gravidez angular – um diagnóstico inesperado

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Abstract

Angular pregnancy is a rare condition characterized by embryo implantation at the angle of the uterus, near the fallopian tube. It has been associated with serious obstetric complications such as fetal growth restriction, placenta accreta spectrum disorders and preterm birth. We report a rare case of a 39-year-old woman with a previous cesarean and a hysteroscopic myomectomy, who was admitted with second-trimester bleeding. Obstetric ultrasound revealed a fundal placenta with two subamniotic haematomas and fetal growth deceleration. A preterm cesarean was performed and when faced with a challenging placenta removal, the diagnosis of an angular pregnancy was established. To our knowledge, few cases describing this condition have been reported. True prevalence may be underestimated because of general lack of awareness and clear criteria for the diagnosis. With this case report we highlight the importance of this diagnosis and its potential complications.

Keywords: Ectopic pregnancy; Angular pregnancy; Bleeding.

Resumo

A gravidez angular é uma entidade rara na qual a implantação do embrião ocorre no ângulo da cavidade uterina, adjacente à junção utero-tubária. Associa-se a diversas complicações obstétricas, nomeadamente restrição do crescimento fetal, acretismo placentário e parto pré-termo. Apresentamos o caso de uma mulher de 39 anos com antecedentes de cesariana e de miomectomia por ressectoscopia, internada por hemorragia do segundo trimestre. A ecografia revelou uma placenta fúndica, dois hematomas subamnióticos e desaceleração do crescimento fetal. Foi realizada uma cesariana pré-termo e, perante uma dequitação difícil, foi realizado o diagnóstico de gravidez angular. Esta é uma situação rara, havendo poucos casos publicados. Este artigo alerta para a importância do diagnóstico e das possíveis complicações obstétricas associadas.

Palavras-chave: Gravidez ectópica, Gravidez angular, Hemorragia.

INTRODUCTION

Angular pregnancy is a rare condition characterized by embryo implantation at the lateral angle of the uterus, near the fallopian tube. Non tubal ectopic pregnancy includes cornual, angular, and interstitial preg-

nancies¹. Although these three concepts are often misused, some differences must be noted. Cornual pregnancy refers to an intrauterine implantation within a uterine horn of an anomalous uterus. In an angular pregnancy, the embryo implants medially to the uterotubal junction whereas in an interstitial pregnancy, the implantation occurs laterally to the utero-tubal junction. Another major difference between these two entities is that interstitial pregnancies are not viable because they never centralize into the uterine cavity¹.

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FIGURE 1. The uterus before placental removal.

Angular pregnancy has been historically associated with serious obstetric complications such as fetal growth restriction, placenta accreta spectrum disorders and preterm birth. The largest published meta-analysis reported 39 cases of angular pregnancy and a 38.5% rate of spontaneous or missed abortion². In a recent case series published by Bollig *et al.*, 42 cases diagnosed in the first trimester scan were surveilled. Adverse outcomes included early pregnancy loss (20%) and preterm birth (18%)³.

CASE DESCRIPTION

A 39-year-old pregnant woman, G2P1 (previous cesarean section due to breech presentation), was admitted at 26 weeks and 5 days of gestation due to second trimester bleeding associated with painless irregular ute-

rine contractions. She had had a previous hysteroscopic myomectomy. There was no history of abdominal trauma. This was the first bleeding episode in the current pregnancy. Ultrasound examination revealed a fundal placenta with two anechoic formations, anterior and posterior to the placenta, compatible with chorioamniotic haematomas. These haematomas seemed to have occurred at different moments and measured 88 x 31 x 95 mm and 54 x 59 x 21mm. During hospitalization, bleeding persisted intermittently for 20 days, although in smaller quantity and without hemodynamic and cardiotocographic repercussions. Frequent ultrasound follow-up was performed, revealing an increase in the size of the haematomas and a deceleration in fetal growth. At admission, the fetus was at the 27th centile and two weeks later at the 7.3 centile (Hadlock curves). Fetal doppler revealed hemodynamic redistribution. The final ultrasound, performed at 29 weeks and 5 days of

gestation, revealed an increase in the size of the haematomas. Doppler indices were as follows: umbilical artery pulsatility index (PI) 1.67 (98th centile), middle cerebral artery PI 1.58 (10th centile), cerebroplacental ratio 0.95, peak systolic velocity 48.1 cm/s, and ductus venosus PI 0.44. Due to these findings and because of the high risk of maternal hemorrhage and fetal death, fetal maturation with corticosteroids and fetal neuroprotection were completed and a cesarean was performed at 29 weeks and 5 days of gestation. The newborn weighed 1006 g and the Apgar score was 7/9/10. Placenta removal was challenging, requiring uterine exteriorization. An asymmetrical bluish bulging area in the right angle of the uterus was identified (Figure 1). After manual placental removal and uterine massage, myometrial contraction was accomplished, and uterine contours became symmetrical. These findings raised the suspicion of an angular pregnancy. There were no incidents to report until discharge, after 4 days of postpartum. Histological examination of the placenta revealed no alterations. One month postpartum, the neonate exhibited favourable clinical evolution. The patient provided informed consent for the publication of this article, including the use of all relevant clinical details and accompanying materials.

DISCUSSION

Angular pregnancy was first described in 1898 by Howard Kelly⁴. This entity has been probably underdiagnosed due to indiscriminate diagnosis of angular, interstitial and cornual pregnancy. It is important to establish the difference between these three entities because they have different prognoses. An interstitial pregnancy will never evolve into a viable pregnancy whereas an angular pregnancy can grow into the uterine cavity and result in a live birth. Angular pregnancy can be evaluated through sequential ultrasound, especially in the first trimester, to confirm its growth into the uterine cavity. In some cases, a 3-D ultrasound may be needed to achieve an accurate diagnosis. At a more advanced gestational age, the diagnosis can be more difficult and should be suspected when the placenta is asymmetrically confined to a specific area of the uterine angle⁵. In our case, the diagnosis was suspected

after birth. Although there were signs of placental insufficiency, these are not specific to this situation. According to Jansen and Elliot in 1981, one of the diagnostic criteria for angular pregnancies is retention of the placenta in the uterine angle after delivery⁶. Regarding the management of angular pregnancy, there is an ongoing dilemma on whether to allow the progression or terminate the pregnancy in view of the previously reported high risk of complications². Close ultrasound follow-up in the first trimester can guide the clinical management of individual cases. In conclusion, the diagnosis of angular pregnancy is challenging but its association with adverse perinatal outcomes urges its recognition. Management of this situation is not consensual, including pregnancy termination or careful follow-up. For this reason, more evidence is needed to guide the clinical management of angular pregnancies.

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AUTHOR CONTRIBUTIONS

Rita Nunes was responsible for drafting the manuscript, while Carlota Cavazza, Andreia Fonseca and Luísa Pinto contributed by critically reviewing and revising the article.

CONFLICT OF INTEREST

Authors declare they have no conflicts of interest.

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