



Delayed Delivery of a Twin Pregnancy and Uterine Atony: Morphological and Clinical Evidence

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This report presents the uncommon case of a 154-day delayed delivery in a spontaneous twin pregnancy associated with uterine atony. After abortion of the first fetus at 16 weeks, a healthy male was born at 38 weeks. Postpartum hemorrhage due to uterine atony, which was successfully treated with prostaglandins, occurred.

■ **Keywords:** twin pregnancy, delayed delivery, uterine atony

In the last ten years, there has been a significant increase in the number of twin pregnancies, particularly in relation to the increased use of assisted reproductive technologies. Concomitantly, the incidence of abortions or of premature births has also increased (Tummers et al., 2003). In the literature, there are reports of twin pregnancies, wherein there is the possibility for expulsion of the first fetus with subsequent continuation of the pregnancy and a positive outcome for the remaining fetus (Wouters et al., 2009). It is in this context that we present a case that occurred in our department of a birth at 38 weeks via cesarean section, followed by uterine atony, after expulsion of the first fetus at 16 weeks.

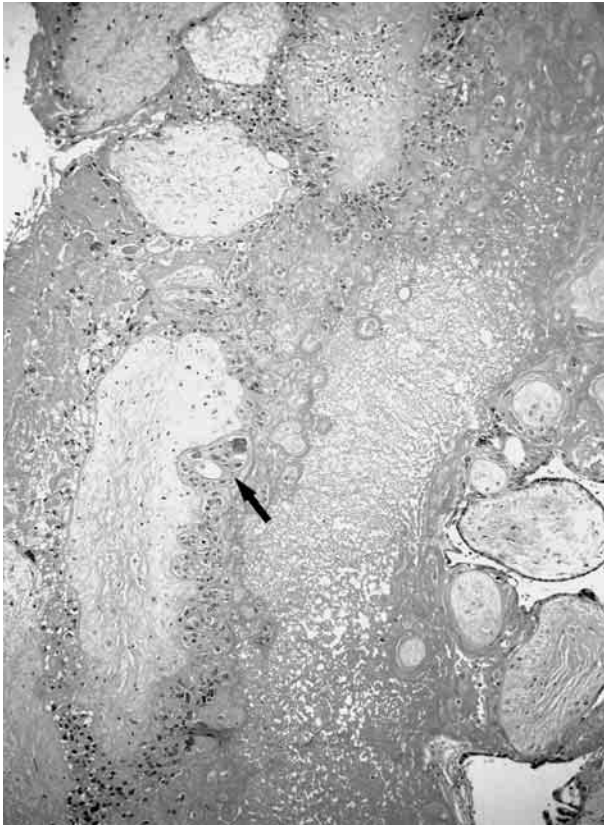
Materials and Methods

A 31-year-old woman, parity 1-0-0-1, conceived a spontaneous twin pregnancy after a prior singleton delivery by cesarean section for fetal macrosomia. At 13 weeks, she was admitted to the hospital due to pelvic pains, metrorrhagia and risk of abortion. Her body temperature, blood pressure and heart rate were normal. The uterine cervix was closed with discreet vaginal hematic loss. Ultrasound observation was performed. Her hematic values were normal, and tocolysis (isoxsuprine hydrochloride, 60 mg/day) was initiated. After four days of hospitalization, the hematic leaks disappeared. At 16 weeks, the first twin spontaneously aborted, and its placenta was retained

within the uterus. After birth of the first twin, tocolysis combined with antibiotics (ceftriaxone sodium, 1 g/day) was continued for 1 month. At 38 weeks, it was decided that hospitalization for extraction of the fetus by an iterative cesarean section would be appropriate. Ultrasound observation was performed. A healthy, male newborn weighing 3,790 g with Apgar scores of 9 and 10 at 1 and 5 min, respectively, was delivered. Subsequently, the placenta of the living fetus (weighing 650 g, regular with respect to form and volume) was easily extracted, while extraction of the atrophic placenta belonging to the fetus expelled at 16 weeks was quite difficult due to its strong attachment to the anterior wall of the uterus. Moreover, postpartum hemorrhage occurred due to atony of the anterior uterine wall, corresponding to the site of the atrophic placenta. Despite the administration of oxytocin (50 IU) and methylergometrine (0.4 mg/M), the atony was only resolved through intravenous administration of sulprostone (0.5 mg). Histological examination of the extracted placentas was performed. The post-partum course of

RECEIVED 28 November, 2009; ACCEPTED 08 December, 2010.

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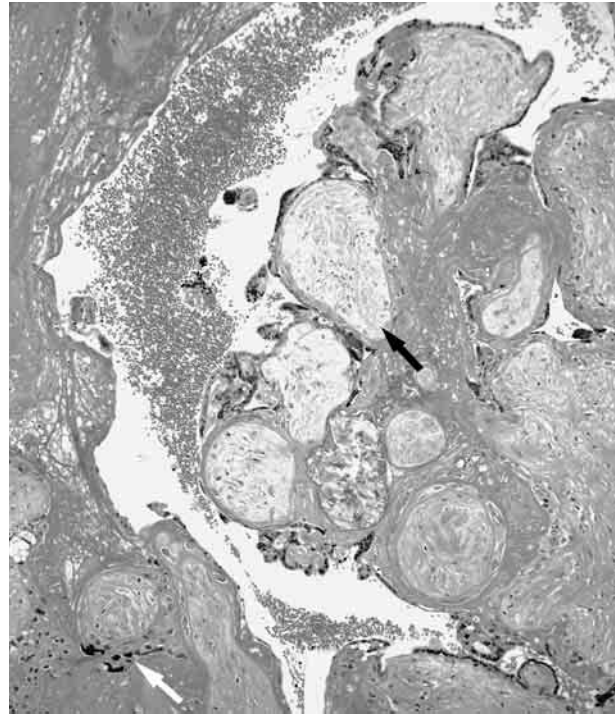
**FIGURE 1**

Chorionic villi show regressive features and necrosis. Surviving trophoblastic cells include isolated syncytial (black arrow) and intermediate-type cells (60x magnification).

mother and infant was trouble-free, and they were discharged on the fifth day after delivery.

Results

The ultrasound scan, performed at 13 weeks, revealed a twin biamniotic and bichorionic pregnancy. One fetus was 25 mm at 9 weeks and lacked cardiac activity inside a dysmorphic gestation area with a detached placental area of 37 for 13 mm (data not shown), whereas the other fetus was alive and was 76 mm, compatible with the period of pregnancy inside a perfectly regular gestational area (data not shown). The ultrasound exam performed at 38 weeks showed fetal growth corresponding to the maximum values for the period of pregnancy and a posterior placenta that was regular with respect to both form and morphology and normal amniotic fluid (data not shown). For the fetus born at 38 weeks, histological examination of the extracted placentas revealed a situation compatible with the secondary placentopathy at vascular district hypertension characterized by arterial-hyperplastic stenosis, with congestion of the villous vessels and confluent hematic gaps. Moreover, focal ischemic necrosis and sclerolinosi in the marginal area were evident, as well as

**FIGURE 2**

Hemorrhagic lacunae can be seen inside the intervillous space near degenerating villi (black arrow) and calcific foci (white arrow) (60 x magnification).

multiple calcifications of the chorionic villus (Figure 1). For the fetus expelled at 16 weeks, there was extended coagulative necrosis and regressive phenomena of the trophoblastic epithelium, with exudative inflammatory infiltrates and micro-calcifications with focal marginal destruction (Figure 2).

Discussion

Women with twin pregnancies are at risk of preterm deliveries that are associated with high perinatal mortality and morbidity. Often, delivery of the first fetus is followed shortly thereafter by delivery of the other twin (Malinowski et al., 2005). However, in this case study, expulsion of the dead fetus at 16 weeks did not cause any alteration in the development of the remaining twin fetus. In fact, the latter twin was born at 38 weeks and was of normal weight. In a literature review of delayed delivery, Wouters et al. (2009) showed that delay of delivery beyond 37 weeks is rare, with only eight known deliveries at term. In six of these eight cases, the first sibling was born before 24 weeks; prolongation appeared to be longer when the first birth was earlier and the interval range between delivery of the first and second twins was 1-153 days. Moreover, Wouters et al. (2009) reported an extraordinarily rare case of delayed delivery after late abortion of the first twin; the observed interval time was 134 days. In our case, expulsion of the first sibling at 16 weeks was followed

by the birth of a healthy male at 38 weeks with an interval time of 154 days. The morphological evaluation of placentas of this twin diamniotic and bichorionic pregnancy enabled the establishment of the type of placenta but not the probable cause of the intrauterine death of the first twin fetus. In a study of twin gestation, Malinowski et al. (2005) used morphological evaluation to determine intrauterine demise during the first trimester. However, the twins in the study were monochorionic diamniotic twins. An undesired event that occurred in our case was postpartum hemorrhage due to uterine atony at the site of the dystrophic placenta of the dead fetus. Despite the administration of oxytocin and methylergometrine, the uterine atony was only successfully treated with prostaglandin therapy based upon recent findings of pharmacological therapy (Breathnach & Geary, 2009). In conclusion, the expulsion of the twin fetus at 16 weeks did not provoke interruption of the pregnancy. To the contrary, the pregnancy lasted an additional 22 weeks with a

rare delay time of 154 days. The only abnormal phenomenon observed was post-partum hemorrhage due to uterine atony, an event that was successfully solved with a recently developed pharmacologic therapy.

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