Late recognition of fetus papyraceus
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Abstract

A 29 year-old gravida 3 para 2 with an IVF dichorionic-diamniotic twin gestation complicated by, a 14 week selective reduction presents with complaints of feeling that something is bulging from her vagina on postpartum day 5. The vaginal pressure worsened and then a malodorous vaginal discharge was noted. On postpartum day 10, the patient presented to the emergency department where an entire mummified fetus, umbilical cord and placenta were removed from the uterus with gentle traction. While fetus papyraceus is a well-described phenomenon in the literature, its late recognition is uncommon. Delayed recognition of fetus papyraceus, or any retained products of conception for that matter, can potentially lead to grave consequences, including disseminated intravascular coagulation and septic shock.

Key words:
Fetus Papyraceus
Selective reduction

Background

“Vanishing twin” is a term given to a single fetus of a multi-fetal gestation that dies in utero and is either partially or completely resorbed. When the fetus becomes mummy-like in appearance and texture, it is often referred to as fetus papyraceus. This is a result of the physical compression by the surviving and growing fetus and the loss of fluid and soft tissue. Risks to the mother and/or surviving fetus include: maternal coagulopathy, preterm delivery of the remaining fetus, neurological abnormalities and fetal demise.

Whether as a result of a spontaneous fetal demise (FD), selective reduction (SR) or selective termination (ST), this phenomenon has become more common since the widespread use of assisted reproductive technology, specifically In-Vitro Fertilization and ovulation induction.

Case

This study presents a case of a 29 year-old, gravida 3, para 2 with an IVF dichorionic-diamniotic twin pregnancy. Prior obstetrical history was significant for a spontaneous abortion of a twin pregnancy at 18 weeks and a preterm vaginal delivery at 32 weeks secondary to preterm labor. This pregnancy was complicated by an elective fetal reduction of one twin at 14 weeks. Close monitoring of the pregnancy
thereafter showed a healthy singleton pregnancy with an appropriately grown fetus.

At 32 weeks gestation, the patient transferred care. The pregnancy was uncomplicated and proceeded with elective induction at 39 weeks. At 39+2 week’s gestation, augmentation of labor resulted in the birth of a healthy female fetus at 6.5lbs. Of note, the umbilical cord was noted to be thin and dull-looking rather than the usual shiny and fat appearance. The third stage of labor was uneventful and blood loss was minimal. The post-partum course was uncomplicated and the patient was discharged home on post-partum day 2.

On postpartum day 6, the patient reported intermediate feelings of vaginal fullness, as if her uterus was prolapsing, that resolved spontaneously. Patient denied any history of abdominal pain, vaginal bleeding or other associated symptoms. An early post-partum visit to her obstetrician and gynecologist was unremarkable, including physical and pelvic exam.

Over the next several days, recurrence of events became more frequent and the symptoms more debilitating. By post-partum day 10, conventional methods at self-reduction of a prolapsing uterus failed and the patient reported a newly developed malodorous smell. The patient was instructed to present to the emergency department for further evaluation. On bimanual exam, the patient was noted to have an approximately 5cm, loosely connected, mass that could be extracted from the vagina. Upon further inspection of this hard-encased mass, bony fragments could be felt; it became apparent that the mass was indeed the remnants of the 14-week reduced twin. Speculum exam enabled the mummified hard remnant to be removed as a single unit, attached to an umbilical cord and placenta, and the specimen was sent to pathology. The patient was put on prophylactic antibiotics and sent home with close follow-up. Pathology confirmed the specimen as fetal remnants — a late diagnosis of fetus papyraceus.

**Comment**

**Prognosis for the surviving fetus(es) of multigestational pregnancies with a fetal demise or selective reduction/termination?**

The prognosis for surviving fetuses of a multigestational pregnancy with a spontaneous fetal demise depends on several factors: number of the fetuses, gestational age at the time of the demise, the reason for the demise, the chorionicity and the length of time between demise and delivery of surviving fetus(es). First trimester demises in multichorionic multifetal gestations will likely result in “vanishing twins” and there doesn’t appear to be any increased risk of complications to the surviving fetus. In cases of selective reduction, there is reported improvement in fetal outcomes. In the largest collaborative multi-center study that looked at outcomes of selective terminations in 402 patients, showed that greater than 90 percent resulted in viable neonates. Late FD or SR/ST increases the risk to the remaining fetus(es). Nonetheless, Evans et al showed that more than 90
percent of an ongoing pregnancy after a selective termination in all trimester’s ended with delivery of viable neonates.\textsuperscript{4}

Management of an ongoing pregnancy after selective reduction/termination.

To date, there is no recommended follow up interval for fetal or maternal monitoring in cases of FD/SR/ST for ongoing pregnancy. Considerations for management should be multifactorial: understanding of cause of death and risk to remaining fetus(es), length of time till term and risk to mother.

Maternal consumptive coagulopathy as a complication of a late fetal demise is a rarely reported complication.\textsuperscript{5,6} A theory for such a low reported incidence of consumptive coagulopathy involves the relative short interval from demise until delivery of the surviving fetus.\textsuperscript{7} “Gross disruption of the maternal coagulation mechanism rarely developed within 1 month after fetal death. Although, if retained longer, approximately 25 percent will develop a coagulopathy.”\textsuperscript{8}

Sanetema et al assessed pregnancy outcome of expectantly managed twin pregnancies in which one of the fetuses died after 20 weeks. They concluded that conservative management outweighed preterm-delivery.\textsuperscript{9}

Conclusion

There is an increase in the incidence of multi-fetal pregnancies as a result of the advances in assisted reproductive technologies. Additionally, with the advanced ultrasound machines and techniques used today, there are even more early multi-fetal gestations being documented. Accordingly, the incidence of a spontaneous vanishing twin or planned SR/ST has risen. Fortunately, improvements in the techniques used for SR/ST have led to improved outcomes for the surviving fetus(es).

Nonetheless, the clinician’s role should include consistent documentation and labeling of gestation status with history of FD/SR/ST, as it is essential to patient safety and ability to provide appropriate medical care. Based on the patients’ medical history and the reason for the fetal FD or SR/ST, patients should be followed closely for fetal wellbeing as well as possibility of maternal infection or consumptive coagulopathy. Patients should receive periodic ultrasounds to assess appropriate fetal growth. Lastly, at the time of the delivery the physician should inspect the placenta for evidence of the “vanishing twin or fetus papyraceus” and if not evident in its entirety, an ultrasound should be performed to document its complete reabsorption or need for surgical management.

Delayed recognition of fetus papyraceus by this magnitude can have grave consequences. Early recognition and prevention is the best cure.


