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Editorial

Management of monoamniotic twins: the question is not 'where?', but 'how?'

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Monoamniotic twins are very rare, with a prevalence of only 8 per 100 000 pregnancies (multiples and singletons combined); they account for fewer than 1% of all twin pregnancies[1]. Their complication rates, on the other hand, are extremely high. Indeed, studies reporting on the outcome of monoamniotic twin pregnancies from the first trimester onwards show them to have significantly higher risks than do dichorionic or diamniotic twin pregnancies, with fetal or neonatal loss rates of up to 20–50%[1–5]. The majority of these losses occur prior to 24 weeks' gestation, but even thereafter, the risk of fetal death remains as high as 5–10%[6], which is double the risk seen in monochorionic diamniotic twins[7].

In contrast to diamniotic twins, only about one third of fetal deaths in monoamniotic twins are due to twin reversed arterial perfusion sequence, twin–twin transfusion syndrome, preterm birth or (pregnancy termination for) congenital anomalies[5,8,9], while the other two thirds are thought to be the result of acute cord occlusion and/or intertwin transfusional imbalances as a consequence of the near-

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universal cord entanglement and the large intertwin placental anastomoses seen in these pregnancies[10–12].

Deaths due to these acute events cannot be prevented prior to fetal viability but preterm delivery becomes a therapeutic option after viability if the adverse event can be predicted in time.

Unfortunately, there is no clear evidence that any method of fetal monitoring can predict accurately fetal death in monoamniotic twins. Notching in the Doppler waveform of the umbilical artery is a very common, and sometimes transient, finding, with poor specificity[13,14]. Similarly, the sensitivity of intermittent fetal heart-rate monitoring is poor, as fetal deaths have been reported to occur just hours after a completely normal cardiotocogram, and continuous fetal heart-rate monitoring is not feasible[15]. Despite this, circumstantial evidence suggests that monoamniotic twins which undergo regular surveillance do better than do those which do not have such surveillance, and fetal death rates have decreased over time. This is in part because most obstetricians now deliver monoamniotic twins by 32–33 weeks at the latest, when the prospective risk of fetal death exceeds the risk of neonatal death or severe morbidity[3], even though Level-1 evidence for this practice is lacking[16]. Additionally, it is likely that some fetal deaths are subacute or have a prodromal phase and can therefore be predicted. As such, extended episodes of fetal tachycardia or repeated heart-rate decelerations , for example, will often be considered as triggers for delivery[17]. This nevertheless leaves us with the clinical questions as to how best to monitor monoamniotic fetuses practically, between the point of viability and the planned Cesarean section; i.e. which monitoring methods should we use, and how often and in what setting should they be applied?

Historically, the first studies attempting to demonstrate a benefit of fetal surveillance in monoamniotic twins compared the outcomes of monoamniotic twin pregnancies admitted to hospital for fetal heart rate monitoring with pregnancies that continued standard outpatient care[18,19]. Both studies showed that inpatients did better than outpatients. Unfortunately, however, this comparison between inpatients and outpatients shifted the focus from the frequency and method of surveillance to the physical location of surveillance, and subsequent studies maintained this polarization. The MONOMONO Working Group study[20], for example, in this issue of the Journal, found that the risk of fetal death was higher in outpatients (10.8%) compared with inpatients (3.3%), although this difference was not statistically significant. However, combing through their Methods section, it quickly becomes apparent that the data do not support such a conclusion, as the location of surveillance was also strongly correlated with the surveillance protocol. The inpatient cohort, besides being admitted to hospital, started fetal surveillance from 24–28 weeks onwards, while the outpatient cohort started surveillance only at 30 weeks' gestation. The vast majority of the excess fetal deaths seen in the outpatient group occurred prior to initiation of intensive surveillance and, when truly comparing apples with apples, analyzing only fetal deaths after 30 weeks (i.e. when both patient groups were actively watched), the incidence of fetal mortality in both groups was very similar: 1.4% in the inpatients and 2.4% in the outpatients (χ^2 test, P = 0.67). Unfortunately, their findings have been integrated into a recent meta-analysis[6], also in this issue of the Journal, which concluded that inpatient surveillance is associated with lower fetal mortality than is outpatient follow-up (5.9% *vs* 14.6%), thereby again confusing physical location with surveillance protocol.

This confusion, although seemingly benign in nature, could have significant implications if it is translated into policy-making. Indeed, in the absence of a clear fetal benefit, hospital admission can be harmful to pregnant women, being associated with an 18-fold increased risk of venous thromboembolism. This risk is even higher in the third trimester and in multiple gestations[21]. Moreover, prolonged hospital admissions incur significant economic and societal costs, and are highly disruptive to family life. In a study of almost 200 women carrying monoamniotic twins, psychiatric symptoms were far more common in the inpatient group compared with in the outpatient group (although the difference did not reach statistical significance), with very high rates of hopelessness and despair (42% *vs* 24%), thoughts of self-harm or suicide (4% *vs* 0%) and postpartum depression (12% *vs* 4%) in the inpatients[22].

It is, therefore, high time that we dissociate the physical location of surveillance from the surveillance protocols used. Despite the heterogeneity of the protocols of the studies performed to date, their reinterpretation suggests that intensive outpatient follow-up, using a combination of fetal heart-rate monitoring and ultrasound, can decrease the risk of fetal death after viability to below 5%[3,8,20]. The difference in fetal survival between daily (formerly called 'inpatient') follow-up and alternate-day (formerly 'outpatient') follow-up, if any, is likely to be small. In our hospitals, therefore, we offer, with clear explanation, both surveillance options and let patients choose their preferred management, after informed consent is obtained.

We hope that future studies will address surveillance protocols rather than just physical location and, as a randomized trial is unlikely due to the rarity of the condition, we encourage all centers to present their results with well-described protocols to allow for more detailed (individual patient) meta-analysis.

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