



Management of Spontaneous Rupture of the Amnion with an Intact Chorion

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Abstract

Idiopathic severe preterm oligohydramnios as a result of spontaneous rupture of the amnion with an intact chorion is a rare event with a scarcity of reports found in the literature. We evaluated the impact of serial amnioinfusions on this unusual occurrence. This is a follow-up of a 37-year-old woman with idiopathic severe oligohydramnios diagnosed at 18 weeks of gestation. We performed five serial amnioinfusions with the purpose of improving fetal lung maturity and to prevent Potter anomalad. At 31 weeks' gestation the patient was delivered of a healthy neonate by classical cesarean section. The child is now 17-years-old and without any physical or mental impairments. This outcome supports the consideration of serial amnioinfusion in oligohydramnios caused by spontaneous rupture of the amnion with an intact chorion. In this rare clinical presentation, amnioinfusion may allow for normal fetal development.

weeks' gestation with an AFI of 1cm. The patient reported no history of vaginal amniotic fluid leakage. Speculum examination showed no evidence of vaginal pooling. The fetal kidneys were visualized at the time of the ultrasound and were considered functioning since the fetal bladder was visualized at the time of ultrasound. Chorionic villus sampling (CVS) was performed after the 18-week scan and showed a normal 46,XX complement.

The patient was informed of the complications accompanied with oligohydramnios and was offered pregnancy termination. When asked if there were any possible therapies, she was informed that serial amnioinfusion could theoretically reduce the morbidity and mortality associated with severe oligohydramnios but that such therapies had not been extensively evaluated and reported in the literature and that the likelihood of a poor neonatal outcome should still be considered high. Nevertheless, she opted to continue the pregnancy with treatment with serial amnioinfusions and informed consent was obtained.

The patient received five transabdominal amnioinfusions between 19 and 25 weeks' gestation. The fluid infused was a warmed (37°C) 0.9 % saline solution instilled under ultrasonographic guidance through a 22-gauge needle into the amniotic cavity. Because of the sensitive phase of lung development, the infusions were commenced soon after detection of oligohydramnios and the decision to continue the pregnancy. The first amnioinfusion was performed at 19 weeks' gestation with 300cc saline and the addition of 5 ml indigo carmine diluted 1:20. A vaginal tampon was used after the procedure for detection of leakage of infused fluid; no blue dye was detected on the tampon. The initial amnioinfusion allowed for a detailed, diagnostic ultrasound for fetal malformations. Sonographic examination disclosed an anatomically normal fetus with normal fetal movements. The second amnioinfusion was performed 7 days after the first one with 400cc saline and 5ml indigo carmine diluted 1:20. There was again no leakage of AF and no anatomical abnormalities detected. 14 days later we again injected 400cc saline. Post infusion the AFI was 13.7cm and 3 days later the AFI was 3.6cm. 12 Days later (AFI: 2,5cm) 450cc saline were injected. The last amnioinfusion 9 days later was performed with 450cc saline. The post AFI was 14.4cm and 3 days later was 8.9cm. A sonographic examination was performed after each amnioinfusion with normal anatomical surveys and normal fetal movements being observed. There was normal fetal

Established Facts

- Oligohydramnios is associated with poor neonatal outcomes, especially if the condition is severe or noted in the second trimester.
- Idiopathic severe preterm oligohydramnios as a result of spontaneous rupture of the amnion with an intact chorion is a rare event, though still associated with severe neonatal morbidity and mortality.
- There is no accepted treatment shown to be effective for salutary short- or long term outcomes in cases of severe oligohydramnios in the second trimester

Novel Insights

- In cases of severe oligohydramnios resulting from rupture of the amnion with an intact chorion, serial amnioinfusion may provide the necessary intraamniotic fluid volume to maintain an ongoing pregnancy

Case Report

The patient is a 37-year-old white female, gravida 3, para 1, unrelated and previously unknown to the treating physician and staff, with an intrauterine IVF pregnancy that was complicated by severe oligohydramnios in the second trimester. She presented at 18

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growth till 29 weeks of gestation (10-25% percentile) and no evidence for IUGR was noted. After each infusion fetal breathing was noted as was increased fetal activity. A separation of amnion from the chorion was identified as a free floating membrane after amnioinfusion. The patient had also been given weekly betamethasone injections from 24 weeks for fetal lung maturity, as was the practice at that time. At 27 3/7 weeks of gestation, the patient presented at labor and delivery with preterm premature rupture of membranes (pPROM), but with no evidence of labor. Clear fluid per vagina was noted with an AFI of 1.6cm, which remained at 2.0cm on a subsequent ultrasound examination. Intravenous ampicillin prophylaxis was started to prevent intraamniotic infection. She remained in the hospital for continued monitoring. At 30 5/7 weeks of gestation the patient had a biophysical profile that was 2 out of 8 and a non reassuring non stress test (NST). Because of the belief that the fetus would not tolerate labor, a classical cesarean was performed without complications. The patient was delivered of a 980gm female fetus [31 weeks small for gestational age (SGA) neonate (<10 % percentile)] with APGAR scores of 8 at 1 minute and 9 at 5 minutes.

The newborn had a normal renal work-up and never demonstrated any respiratory disease. There was normal physical development and no physical malformations. Eight weeks after delivery the healthy baby was discharged from the hospital. That neonate is now a 17-year-old female (height 5 ft 1 in, weight 94lbs), and is an A-student in high school and a part-time working model, with no apparent physical or mental deficiencies.

Discussion

Second trimester oligohydramnios is a predictor of poor fetal outcome and often an indication for pregnancy termination, regardless of the cause of the oligohydramnios [1-3]. Barss et al. described 12 cases with severe oligohydramnios in the second trimester with a mortality rate of 100%. Severe oligohydramnios can result in an impairment of the ultrasonographic acoustic window; accordingly, fetal anomalies may be missed by ultrasound examination. Amnioinfusion may thus improve the detection of fetal anomalies as well as the outcome in premature pregnancies complicated by oligohydramnios [4-7].

The most likely complications of amnioinfusion are amniorrhexis and infection. In the study from Fisk et al. [4], only 2 of 89 patients (2.2%) developed amnionitis after amnioinfusion and Gramellini et al. [8] also concluded that antepartum amnioinfusion seemed to offer more benefits than negative effects. In preliminary publications, amnioinfusion has been suggested as an intervention that can improve perinatal outcome and prolong pregnancies, especially in cases of pPROM [6]. Spontaneous rupture of the amnion with an intact chorion is a extremely rare event with no other case report found in literature. This is the most likely etiology for those reported idiopathic cases. An intact chorion prevents external leakage but doesn't prevent maternal reabsorption. There is little known about successful management of this rare occurrence and even less information about long term follow-up.

As such, few reports in the literature are readily comparable to our case. Fisk and colleagues performed serial saline amnioinfusions in 3 idiopathic cases with an onset of oligohydramnios between 11 and 22 weeks of gestation. The mean gestational age at the beginning of the infusions was 20-23 weeks. Two neonates (66%) survived and one neonate died because of respiratory distress syndrome. In a current review Kozinszky et al. (2014) [9] summarized all available cases so far concerning treatment of severe midtrimester oligohydramnios. Out of 289 pregnancies were treated with serial amnioinfusions; only 34 cases (11%) were idiopathic with a mean gestational age at diagnosis of 21.7 weeks. Ten idiopathic cases treated with saline amnioinfusions had a 50% perinatal survival rate with no information about fetal outcome. Chen et al. [10] treated 7 idiopathic cases with serial Ringer's lactate amnioinfusions. Fifty-seven percent survived, with no evidence of fetal abnormalities. Kozinsky et al. (2013) [11] performed 17 saline amnioinfusions for idiopathic oligohydramnios; the survival rate was 35%. In the period from 1986-

2014 (28 years) there were just 37 cases of idiopathic severe second trimester oligohydramnios from the literature. Adding our case to the 37 additional cases, results in an overall survival rate of 47% after treatment with serial amnioinfusions. The average latency from the first multiple amnioinfusion till delivery was 52.6 days. This confirms that an increase of AF volume by invasive treatment results in over 400% greater improvement in fetal outcome, compared with the high mortality rate in cases without amnioinfusion [2,3]. Pulmonary hypoplasia, the most common cause of neonatal death, may be predicted by the lack of fetal breathing movements. Blott et al. [12] showed that all cases with PROM in the second trimester and fetal breathing were associated with a favourable fetal outcome. In our study fetal breathing was noted after each amnioinfusion. Based on our results, we suggest that normal fetal breathing movements can be used as a predictor for normal lung development.

We therefore recommend that in cases of idiopathic oligohydramnios characterized by normal karyotype and fetal anatomy, the offering of pregnancy continuation with serial amnioinfusions as an option for pregnancy management. This case demonstrates that serial amnioinfusion can result in a viable pregnancy outcome, and allow for normal development and a satisfactory long term postnatal outcome. We hope that this report will thus encourage others to report outcomes of cases employing this management strategy.

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