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Case Report

Multiple Fetal Skeletal Deformations and Uterine Wall Distortion; Two Rare Complications of Prolonged Preterm Premature Rupture of Membrane

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Abstract

We present a rare case of multiple fetal skeletal-deformations and uterine-wall distortion in the presence of prolonged preterm premature rupture of membrane (P-PROM).

A 44-year-old nullipara with a history of unexplained/age-related infertility was admitted at 23^5 gestational weeks due to P-PROM. The patient chose expectant management and at the age of 30^3 gestational weeks, due to fetal-heart decelerations, transverse fetal-lie, she underwent a classical cesarean-delivery. At inspection of the newborn there were multiple skeletal deformities; an asymmetrical chest wall, asymmetrical skeletal head, distortion of right arm and mal-position of right ankle. X-ray showed a fishtail deformity of the right distal humerus. Upon revision of the uterus, to our surprise, given a normal uterine cavity on hysteroscopy one month prior to pregnancy, a thick oblique septum-like structure was observed, attached to the back, fundal and right uterine-walls. Part of the placenta was attached to the right side of the septum. The structure was excised, enabling to reach a single uterine cavity. Pathology revealed right side of septum with decidua and left side compatible with uterine-wall. It is possible that lack of amniotic fluid and with it decreased positive pressure on uterine-wall facilitated the detachment of the left side from the uterine-wall resulting in a deformed uterine-cavity. The newborn was admitted to the neonatal intensive care unit. During her admission she developed mild RDS, received one dose of surfactant and at the age of 46 days she was free of respiratory support. She was discharged at the age of 65 days, with mild developmental dysplasia-of-hip necessitating a splint.

Diagnosing a normal uterine cavity by hysteroscopy could be misleading. Distorted uterine-cavity may enhance the risk for fetal skeletal-deformation in the presence of prolonged-PPROM.

Keywords: *Preterm Premature Rupture of Membrane; Fetal Skeletal Deformation; Uterine Cavity*

Introduction

We present a rare case of multiple fetal skeletal-deformations and uterine-wall distortion in the presence of prolonged preterm premature rupture of membrane (P-PROM).

Case Report

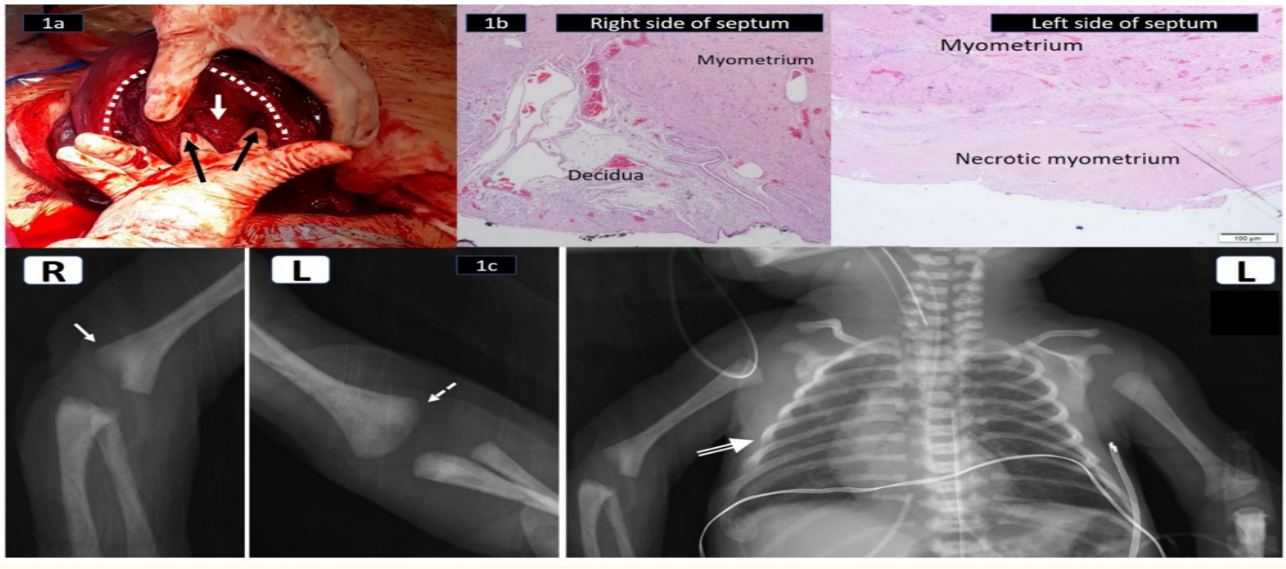
A 44-year-old nullipara G3P0AB1Ect1, was referred at 23^5 gestational weeks due to P-PROM. Her medical background was uneventful apart for 11 years of unexplained/age-related infertility. As part of her infertility evaluation, she underwent hysterosalpingogram (suspected of intracavitary-synechia), repetitive ultrasonogram (suspicious of a small-fundal septum) and 4 diagnostic hysteroscopies (all imaged a normal uterine cavity). Current pregnancy was achieved after nine cycles of in-vitro fertility [1-5].

As fetal survival in Israel is defined at 24^0 gestational weeks, parents are eligible to elect termination of pregnancy if P-PROM occurs prior. After a thorough consultation describing potential maternal and neonatal complications the parents chose expectant management. She was admitted, treated with the Mercer P-PROM protocol and received steroids for lung maturation [1]. Screening for chorioamnionitis included daily fever measurements and complete-blood-count twice-weekly. Fetal wellbeing was followed by fetal heart-rate-monitoring twice-daily and biophysical-profile twice-weekly [1]. Noteworthy, reduced amniotic-fluid-index (< 15 mm) was documented through-out the hospitalization.

At 30^1 gestational weeks an equivocal monitor was documented. She received rescue-dose steroids, magnesium-sulfate (neuroprotection) and at 30^3 gestational weeks, due to fetal-heart decelerations, transverse fetal-lie, she underwent a classical cesarean-delivery.

Upon revision of the uterus, a thick oblique septum-like structure was observed, attached to the back, fundal and right uterine-walls. Two compartments on both sides of "septum" were exposed with only the left one continuous to the cervix (Picture 1a). Part of the placenta was attached to the right side of the septum. The structure was excised, enabling to reach a single uterine cavity. Pathology revealed right side of septum with decidua and left side compatible with uterine-wall (Picture 1b).

At inspection of the newborn there were multiple skeletal deformities; an asymmetrical chest wall, asymmetrical skeletal head, distortion of right arm and mal-position of right ankle. X-ray showed a fishtail deformity of the right distal humerus (Picture 1c). The newborn was admitted to the neonatal intensive care unit. During her admission she developed mild RDS, received one dose of surfactant and at the age of 46 days she was free of respiratory support. She was discharged at the age of 65 days, with mild developmental dysplasia-of-hip necessitating a splint.



1a. A classical uterine incision (dotted white line) revealed a thick oblique septum (white arrow), attached to the back, fundal and right uterine walls. Two compartments on both sides of septum were exposed (black arrows) with only the left one continuous to the cervix.
1b. Histopathology of both sides of excised septum : right side showing the decidua and left side revealing necrotic uterine myometrium. This finding implicates that the left side was initially attached to the uterine wall, lack of amniotic fluid and with it decreased pressure facilitated the detachment of the left side from the uterine wall resulting in a deformed uterine cavity.
1c. Lateral x-rays of the elbows demonstrate hypoplastic right distal humerus (white arrow) and normal left distal humerus (dashed white arrow). Chest radiograph demonstrate bell-shaped thorax with ribs asymmetry and mediastinal shift to the right(white double broad arrow).

Picture 1

Discussion

P-PROM near the limit of fetal viability is estimated to complicate 1:250 pregnancies and the median time for delivery is 3 weeks [2]. Common neonatal complications include: fetal loss, chorioamnionitis, pulmonary hypoplasia and complications of extreme prematurity among surviving infants [3]. Skeletal deformities in the setting of mid-trimester P-PROM are rare. Reviewing 79 surviving newborns with mid trimester P-PROM [4,5] reported only 2 cases of skeletal deformities and contractures. In the current case, fetal movement was restricted due-to the combination of distorted uterine cavity and reduced amniotic fluid. The distorted uterine cavity with the septum like structure was surprising; given that all four hysteroscopies, including the one performed one month prior to current pregnancy, were all normal. Histopathology of both sides of excised septum; right side showing the decidua and left side revealing necrotic uterine myometrium implicate that the left side was initially attached to the uterine-wall. We assume that lack of amniotic fluid and with it decreased positive pressure on uterine-wall facilitated the detachment of the left side from the uterine-wall resulting in a deformed uterine-cavity. In the case presented, the patient underwent hysterosalpingogram suspected of intracavitary-synechia and repetitive ultrasonogram suspicious of a small-fundal septum yet repetitive hysteroscopy imaged a normal uterine cavity. Retrospective, it seems that there was an intracavity synechia or a small septum, which were misdiagnosed by the hysteroscopy (positive water pressure).

Conclusion

Diagnosing a normal uterine cavity by hysteroscopy could be misleading. Distorted uterine-cavity may enhance the risk for fetal skeletal-deformation in the presence of prolonged-PPROM.

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None.

Conflict of Interests

There is no conflict of interest.

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