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Isolated Fetal Pleural Effusion with Progression to Non-Immune Hydrops Fetalis: A Case Report and Literature Review

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Case Presentation

Fetal pleural effusions are fluid collections in the chest cavity of a developing fetus. Pleural effusions can be characterized into primary and secondary etiologies. Primary pleural effusions usually result from lymphatic malformation and are unilateral and isolated findings. Primary pleural effusions are more common in males to females with a 2 to 1 ratio [1]. Secondary pleural effusions are usually associated with structural or infectious etiologies and sonographic, genetic, and infectious work up is necessary as primary pleural effusion is a diagnosis of exclusion [2].

Most often small pleural effusions will regress spontaneously or stabilize, allowing for conservative surveillance and management of such patients [3]. Some may progress to the contralateral side and can potentially lead to development of non-immune hydrops fetalis (NIHF), a serious obstetrical complication diagnosed by the presence of two or more abnormal fluid collections in the fetus, including ascites, pleural effusions, pericardial effusion, and/or skin edema [4]. The most common cause is cardiovascular in origin, however, it is often multifactorial and can involve thoracic obstruction, arrythmias, infections, hepatic venous congestion and/or anemia [2]. The development of hydrops is dependent on etiology, but usually has a poor prognosis with risk of intrauterine fetal demise (IUFD) [5].

The purpose of this case report is to describe the diagnosis and evolution of a patient with fetal pleural effusion with eventual progression to NIHF with a good perinatal outcome.

A 26-year-old primigravid female at 28.4 weeks gestation with resolved COVID-19 infection from 2 months prior presented for consult for concern of fetal ascites on recent ultrasound. Prior ultrasounds revealed a normal anatomy at 20 weeks and an estimated fetal growth of 343grams (75%tile). Prenatal records revealed maternal blood type B, Rh positive, HIV, RPR nonreactive, Varicella, Rubella, Parvovirus immune and cell free DNA screening that was low risk and consistent with a male.

On detailed ultrasound in the high-risk unit, the fetus had an isolated moderate size right pleural effusion with otherwise normal anatomy (Figure 1). Fetal echocardiogram revealed moderate to large right pleural effusion again with normal cardiac anatomy, rhythm, and function.

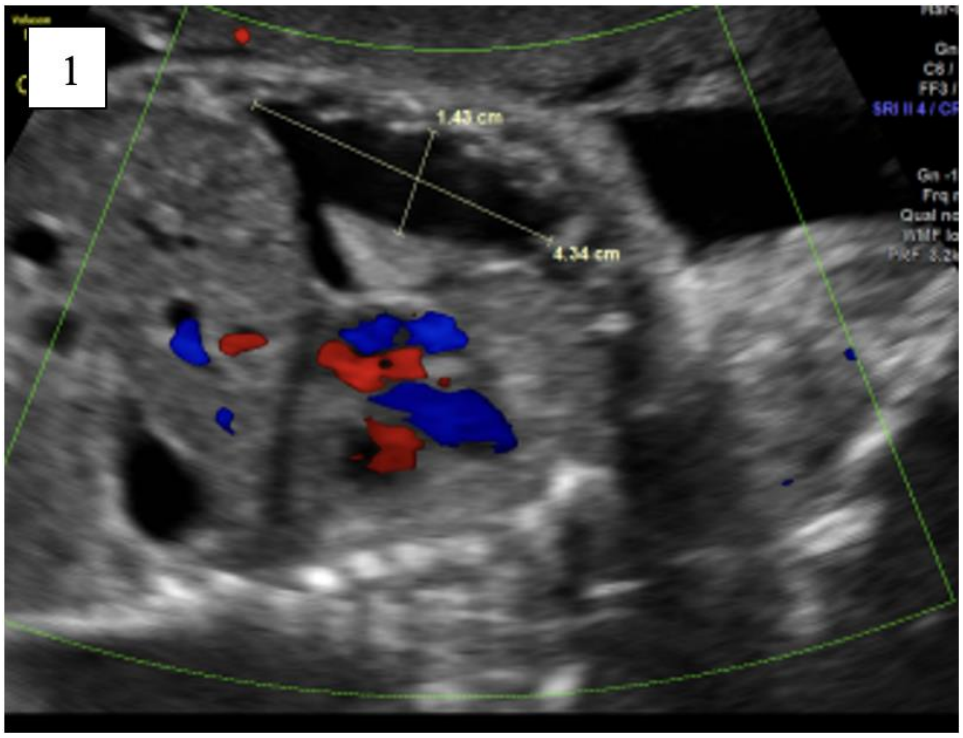

Figure 1: Initial ultrasound at 28 weeks gestation with right sided pleural effusion measuring 4.3x1.3x1.0cm^3.

Weekly ultrasound surveillance found progressive polyhydramnios (AFI 26.2cm to 46.1cm) but stable pleural effusion (7.2x5.7x2.4cm^3) and reassuring peak systolic velocity on middle cerebral artery imaging revealing no indirect indications of fetal anemia (Figure 2 and 3). Skin edema developed at 31 weeks and betamethasone was administered given the progression to NIHF (Figure 4). A genetics consult was placed, and an amniocentesis was offered, which she declined. A thoracocentesis was offered which the patient was amenable to and scheduled. However prior to scheduled procedure, the patient presented to Labor and Delivery with spontaneous preterm prelabor rupture of membranes at 32 weeks and 4 days gestation and was found to be 1.5cm dilated. Latency antibiotics were started. She continued to make cervical change, and a male fetus was delivered at 32weeks and 6 days gestation via primary cesarean delivery with Apgars 4 at 1 minute, 6 at 5 minutes and 8 at 10 minutes and weight of 2450 grams.

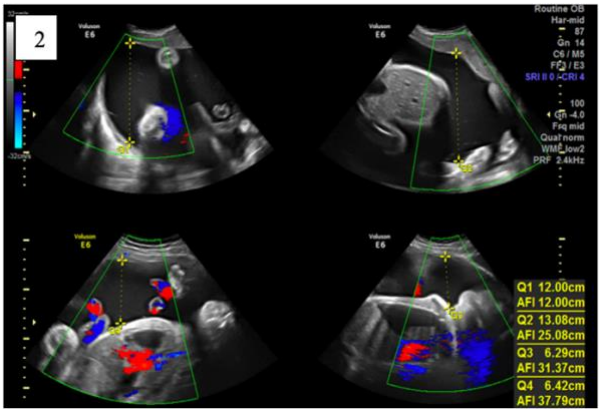

Figure 2: Ultrasound at 31 weeks' gestation with increasing polyhydramnios of 37.8cm.

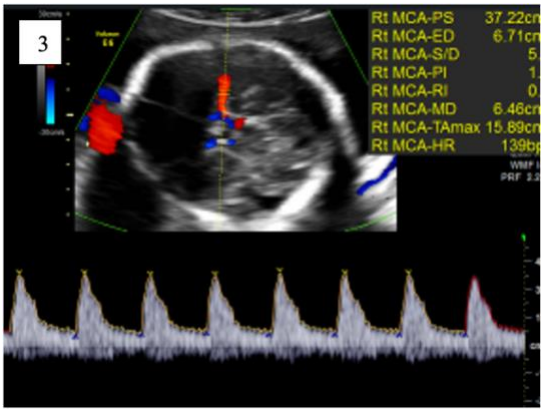

Figure 3: Ultrasound at 31 weeks' gestation with normal peak systolic velocity on middle cerebral artery dopplers.

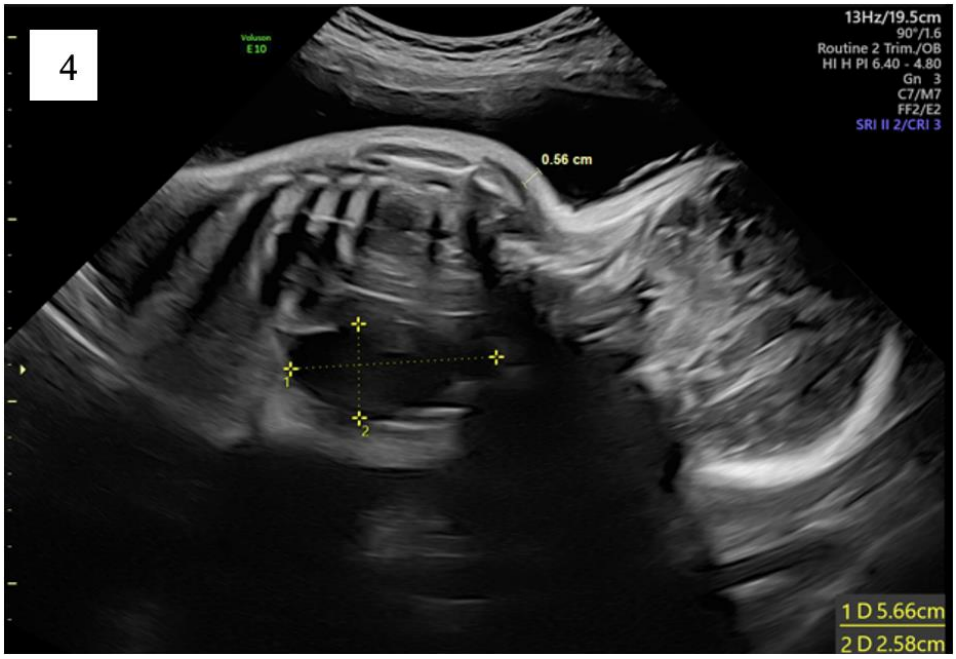

Figure 4: Ultrasound at 31 weeks gestation revealing development of skin edema measuring 0.56cm.

The baby was intubated and transferred to neonatal intensive care unit (NICU). Postnatal chromosome analysis revealed a normal male karyotype. He tested negative for COVID-19. The infant was then extubated, and status post an octreotide drip and chest tubes with no further pleural fluid accumulation. He had video-assisted thoracoscopic surgery and pleurodesis with lung biopsy revealing acute/subacute lung injury and acute fibrinous pleuritis. At 5 weeks old, the current diagnosis was congenital chylothorax verses lymphangiectasia, as initial fluid analysis from pleural effusion revealed 100% lymphocytes.

Isolated fluid collections can be challenging, especially given the unpredictability of diagnosis and progression6. In the case presented, the progression of disease from unilateral pleural effusion to polyhydramnios and skin edema consistent with NIHF suggests a secondary cause. Antenatal etiology was unclear with negative workup for chromosomal, infectious, or structural causes. Drainage of large pleural effusions prior to delivery is recommended for both therapeutic and diagnostic reasons [4]. The finding of lymphocyte predominant pleural fluid may have aided in earlier diagnosis of chylothorax.

The survival and good prognosis of this case can be attributed to a few factors. First, the lack of fetal chromosomal or obvious structural anomalies on ultrasound has been associated with better outcomes [7]. Second, late onset of hydrops is a good prognostic factor. Mcoy et al. found that fetuses diagnosed with hydrops before 24 weeks' gestation were more likely to have abnormal karyotypes and have a perinatal mortality of 95% [8]. Third, NIHF with polyhydramnios has been shown to have a lower risk of IUFD but a higher risk of preterm birth [5]. The increased risk of preterm labor that is conferred with polyhydramnios allude to the possible benefit for earlier delivery for patients with NIHF. Further investigation on timing from diagnosis to fetal morbidity and mortality can help guide delivery timing to balance prematurity and risk of IUFD. There is also limited data on fetal long-term outcomes and should be explored to guide treatment and patient counseling.

It is also important to note the patient's COVID-19 infection shortly prior to diagnosis of fetal pleural effusion. There have been cases of newborns developing pleural effusions secondary to COVID-19 myocarditis or pneumonia [9,10]. This infant had a negative COVID test, and the current diagnosis is related more to lymphatic etiology. However, continuing to understand the effects of COVID-19 in-utero can help elucidate what role this infection may have in fetal pleural effusions and ultimately NIHF.

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