

Spontaneous Resolution of Idiopathic Hydrops in One Twin of a Dichorionic Twin Pregnancy

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Abstract: *Hydrops Fetalis can be due to immune or a variety of non-immune causes. Systematic evaluation of hydrops fetalis helps in finding the aetiology though it can be idiopathic in about one in five cases. Prognosis is generally poor though it varies based on the cause and select in utero treatments can help in improving the outcomes. Very rarely spontaneous resolution can happen. This usually is seen with parvo infections and sometimes with a few structural abnormalities. In some instances, resolution happens in idiopathic hydrops. In Dichorionic twin pregnancy, the cause of hydrops is like that of a singleton pregnancy. Here we report the case history of a Dichorionic twin pregnancy where hydrops was noted in one of the twins at 20 weeks –possibly of idiopathic origin and spontaneous resolution occurred with good outcome.*

Keywords: hydrops fetalis, dichorionic twins, idiopathic, resolution

1. Introduction

Hydrops Fetalis can be immune or non-immune and can be due to diverse etiological causes. The prognosis is usually guarded but varies based on the cause. Here we present the case report of a Dichorionic Diamniotic (DCDA) twin pregnancy in which one of the twin developed hydrops at 20 weeks and describe the pregnancy course and outcome.

2. Case Summary

34 years old G3P2L0 booked at nine weeks of gestation with DCDA twin gestation. It was a spontaneous conception. The patient has had two previous pregnancies with bad outcomes. In her first pregnancy she developed severe preeclampsia and unfortunately had an Intrauterine foetal demise (IUID) at 32 weeks. In the second pregnancy, she had a caesarean section at 34 weeks for preterm premature rupture of membranes but had a neonatal death at three days of age due to Respiratory Distress Syndrome.

In the current pregnancy, all her booking investigations were normal, and she was started on folic acid and low dose aspirin. FTS (First trimester screening) scan was performed at 12 weeks and was normal. Detailed anomaly scan performed at 21 weeks of gestation showed that foetus A had mild pleural effusion and ascites and therefore was diagnosed with hydrops fetalis. Rest of anatomical survey of foetus A and foetus B was normal with no obvious abnormalities detected. There was no growth discrepancy and dopplers including Middle Cerebral Artery Pulsatility Index (MCA) was normal for both foetuses. Possible causes of foetal hydrops were investigated. Both mother and her partner were Rhesus positive. There was no history of any febrile illness in the recent past. Maternal blood pressure, Glucose tolerance test, thyroid function, Haemoglobin and MCV (Mean Corpuscular volume) were normal. Couple were counselled about the diagnosis and the need for invasive testing, but they declined any investigations or

interventions. They were counselled regarding the guarded prognosis and were advised close follow up.

Repeat scan was performed in two weeks to look for progression of hydrops and further repeat detailed anatomical survey to look for cardiac anomalies, thoracic abnormalities, foetal tumours, skeletal defects, markers of aneuploidy, growth discrepancy between foetuses and foetal cardiac rhythm disorders. All the above and foetal echocardiography and Doppler parameters were normal.

Further scan in two weeks showed similar findings with no progression of hydrops. The follow up scan performed at 28 weeks surprisingly showed resolution of pleural effusion and at 34 weeks complete resolution of hydrops of foetus A was noted. The interval growth and Amniotic Fluid Index were normal with no growth discrepancy. Meanwhile the patient developed gestational hypertension and thrombocytopenia. There was no proteinuria and liver enzymes were normal. She was closely monitored and LSCS was performed at 35 weeks for possible HELLP syndrome and previous LSCS. Two healthy female foetuses of 1.84 kg and 1.94 kg were delivered with good APGAR scores. Babies were admitted to Neonatal Intensive Care Unit for observation where the stay was uneventful and were discharged at day 20 of life. Post-natal follow up of babies at 3 months and 5 months have been normal.

3. Discussion

Hydrops fetalis is recognised by the collection of fluid in two or more body cavities-abdomen, thorax or skin. Immune hydrops is due to maternal sensitisation to foetal red blood cell antigen and subsequent transplacental transfer of antibodies resulting in destruction of red blood cells and anaemia. It is most seen with rhesus isoimmunisation but can also be because of other maternal red cell antibodies. Nonimmune hydrops can be due to a variety of causes such as chromosomal abnormalities, infections, haematological

conditions, structural abnormalities, metabolic and genetic conditions [Désilets et al., 2013]. Hydrops in twins is most seen in Twin to twin transfusion syndrome in monochorionic twin pregnancies. In dichorionic twins the causes are like that in a singleton pregnancy.

Our patient had a twin pregnancy and as it was dichorionic, the evaluation will be like that in singletons. However, the patient declined all further investigations. As she was rhesus positive, rhesus isoimmunisation is ruled out. As the MCA dopplers were normal, it is unlikely that there was anaemia and so rules out immune hydrops due to other red cell antibodies as well. Maternal infection screening for toxoplasmosis, rubella, cytomegalovirus, parvo (TORCH panel) would have been helpful but again absence of anaemia rules out parvo virus and there were no other markers of infection on scan. Thalassemia is very common in Asian population but the maternal MCV was normal and no anaemia and so again this is unlikely to be the cause. There were no obvious structural abnormalities noted on the scan. So, the aetiology is possibly idiopathic or due to rare genetic or metabolic causes. A thorough systematic evaluation is important to arrive at the aetiology, though in around 17% of cases the cause cannot be found and is probably idiopathic in origin (Désilets et al., 2013).

The prognosis of hydrops especially nonimmune is poor. There are a few treatment options available in specific situations like intra uterine transfusion and interventions like shunting and drug treatment which can help in improving the outcomes. Society of Maternal and Foetal Medicine-SFM recommends TOP in early onset hydrops (before the viability) (Norton et al., 2015). The couple were counselled regarding the guarded prognosis but opted to continue the pregnancy. She was followed up with regular scans to monitor the foetal conditions.

Spontaneous resolution of hydrops is very rare. There have been a few case reports of spontaneous resolution due to parvo infection and occasionally due to structural abnormalities that have been reported in literature. Also, resolution has been reported in a few cases of hydrops of idiopathic aetiology. Henrich et al had reported on a similar case with hydrops noted at 20 weeks that resolved spontaneously (2002). Iskaros et al had discussed the outcome with early onset hydrops before 18 weeks and noted spontaneous resolution in a few fetuses with normal karyotype (1997). Moreno et al also discussed about spontaneous resolution in a few cases of idiopathic hydrops (2013). Swain et al discussed about spontaneous resolution in two cases of idiopathic hydrops [(1999). Kirkinen et al had reported on two cases of spontaneous resolution (1987). However, all these cases have been in association with singleton pregnancies only.

Our patient had hydrops in one of the twins at 20 weeks. The patient declined any further workup and it was surprising to note the spontaneous resolution of hydrops. The babies were born healthy with no evidence of any abnormalities and remained well at 5 months follow up. Though hydrops in a DCDA twin pregnancy has similar aetiology and course like singletons, we could not identify any reported cases on spontaneous resolution in one twin of a dichorionic twin

pregnancy. In general, hydrops carries a poor prognosis. However, in a few cases of idiopathic hydrops, spontaneous resolution can happen and so conservative management with monitoring can be considered. It is important that patient counselling is done clearly to avoid unrealistic expectations.

4. Conclusions

In general, nonimmune hydrops fetalis-NIHF carries a poor prognosis. Thorough evaluation for the possible causes is important and though rare, spontaneous resolution can happen in a few patients.

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