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Case Series

Use of Antioxidants in the Treatment of Neonatal Hemochromatosis: Two Short Reports

Nada Boutrid[1,2], Hala Boutrid[3] and Hakim Rahmoune[1,2]*

[1]Department of Pediatrics, University Hospital of Setif, Setif 1 University, Algeria

[2]LMCVGN Research Laboratory, Setif-1 University, Algeria

[3]Department of Obstetrics-Gynecology, University Hospital of Bab-El-Oued, Algiers1 University, Algeria

***Corresponding Author:** Hakim Rahmoune, LMCVGN Research Laboratory and Department of Pediatrics, University Hospital of Setif, Setif-1 University, Algeria.

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Abstract

Neonatal hemochromatosis is the most common cause of neonatal liver failure and the leading indication for liver transplantation in infants, characterized by progressive iron deposition during.

It refers to the clinical diagnosis of hepatic failure frequently recognized to be a congenital alloimmune hepatitis, and depicts a severe neonatal liver disease with extrahepatic siderosis, i.e. progressive iron deposition during the fetal period, predominantly targeting the liver, pancreas, heart, and thyroid and salivary glands that spares reticuloendothelial system.

Identifying the characteristic presentation of neonates with neonatal hemochromatosis may help to outline different management strategies for infants, such as antioxidants.

We present two instructive case reports with favorable outcome after supportive care and Vitamin E with N-acetyl-cysteine and clinical presentation of neonatal hemochromatosis is reviewed, along with different treatment strategies, including antioxidant therapy.

Keywords: Neonatal Hemochromatosis; Liver Failure; Ferritin; Alloimmune; Antioxidants

Introduction

Neonatal hemochromatosis (NH) is the most common cause of acute liver failure in the neonatal period; it is associated with a high morbidity and mortality and is due to iron overload in hepatic and extra-hepatic tissues.

Gathering laboratory results to history and clinical examination associated to high ferritinemia allow decisive therapies, including Antioxidants.

Case Presentations

First case

A 45-days-old girl presented pallor, fatigue and failure to thrive; laboratory tests revealed acute hepatitis with liver failure. Alpha-fetoprotein (AFP) was 64,000 IU and serum ferritin > 2000 ng/ml. The most likely diagnosis, NH, required to initiate combination therapy with N-acetylcysteine and vitamin E. After two months of treatment, there were no more stigmata of liver failure; AFP was 3200 IU and ferritin = 632 ng/ml.

The patient was referred for specialized clinics and the family was counselled regarding the possible prophylactic use of immunoglobulins during eventual further pregnancies.

Second case

A 1600-g female infant was delivered at 37 weeks' gestational age by a 30-year-old primiparous mother via an emergent cesarean section for preeclampsia. The infant had normal Apgar scores and

was well till day 10 when she presented anemia, jaundice and global hypotonia.

Investigations were close to the first case along with moderate coagulopathy (prolonged partial thromboplastin time).

The patient was admitted in the neonatal department and recovered progressively after 15 days of vitamin E with N-acetylcysteine and supportive therapy.

During the follow-up period, her laboratory results were normalized within three months.

The case was re-explained to parents, as well as the risk of recurrence and the potential treatment.

Discussion

NH is the most common cause of neonatal liver failure and the leading indication for liver transplantation in infants [1,2].

It refers to the clinical diagnosis of hepatic failure that is largely caused by gestational alloimmune liver disease, recognized to be a congenital alloimmune hepatitis and is defined as the association of severe neonatal liver disease with extrahepatic siderosis, i.e. progressive iron deposition during the fetal period, predominantly targeting the liver, pancreas, heart, and thyroid and salivary glands that spares reticuloendothelial system [1,2].

Other causes of NH encompass perinatal infection, trisomy 21, metabolic disorders and inborn errors of metabolism, and some particular syndromes [1].

Identifying the characteristic presentation of neonates with NH may help to outline different management strategies for infants, such as antioxidants [3-6].

Among antioxidants, N-acetylcysteine is considered as an effective precursor of cysteine for glutathione synthesis and thus, it is widely used as powerful antioxidant *in vivo* and *in vitro*, as it acts directly as a scavenger of free radicals, especially oxygen radicals. Its strength is targeted replenishment of glutathione in deficient cells, like in injured hepatocytes [7].

Currently, alloimmune forms benefit from intravenous immunoglobulins and eventually from exchange transfusion and these successful therapies are widely reported to be efficient and safe [8].

Finally, the diagnosis of NH has prompt implications for future conceptions: recent guidelines support the use of intravenous immunoglobulins during ulterior pregnancies in previously affected families to avoid such severe neonatal hepatic involvement [9].

Conclusion

Hepatic failure in a newborn infant should raise the suspicion of neonatal hemochromatosis.

Antioxidants may offer a window of opportunity, while immunoglobulins seem to be the cornerstone treatment in alloimmune forms, as well as the best preventive.

Ethics Statement

Full parental consents previously obtained for the purpose of this study.

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Conflicts of Interest Statement

The authors have no conflicts of interest to disclose.

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