

Recurrent Intrahepatic Cholestasis of Pregnancy with History of Fetal Demise

Zelalem Mulu^{1,2,*}, Abdulsemed Nur^{2,*}, Hiwot Belachew^{2,*}, Wubshet Assefa^{3,*}

¹Department of Internal Medicine, School of Medicine, Debre Markos University, Debre Markos, Ethiopia; ²Department of Gastroenterology and Hepatology, School of Medicine, Addis Ababa University, Addis Ababa, Ethiopia; ³Department of Pathology, school of medicine, Debre Markos University, Debre Markos, Ethiopia

*These authors contributed equally to this work

Correspondence: Zelalem Mulu, Email zmlaew@gmail.com

Abstract: Intrahepatic cholestasis of pregnancy (ICP) is a rare, potentially fatal liver disorder that occurs in late pregnancy. It is characterized by pruritus, jaundice, and elevated liver enzymes, with spontaneous relief of signs and symptoms after birth. Early detection and treatment are required due to the potential risk of fetal complications, including fetal death. A 30-year-old woman with deeply icteric sclera and skin scratch marks presented to a gastroenterology–hepatology referral clinic. Her liver enzymes, bilirubin levels, and serum bile acid levels were all abnormally high. She also disclosed similar symptoms in her previous pregnancy, which resulted in fetal death at the sixth month. With a presumptive diagnosis of recurrent ICP, the patient was started on ursodeoxycholic acid (UDCA) pills, which significantly improved her pruritus and returned her bile acid levels to normalcy after 2 months of treatment. The delivery was uneventful. We believe that a proper diagnosis combined with UDCA treatment and vigilant obstetric follow-up significantly reduced the patient's symptoms and prevented a possible intrauterine death.

Keywords: pruritis, jaundice, prurigo gravidarum, obstetric cholestasis

Introduction

Intrahepatic cholestasis of pregnancy (ICP) is a potentially serious liver disorder that occurs in late pregnancy. It is diagnosed by the presence of pruritus with onset in late pregnancy, elevated bile acid levels, and relief of signs and symptoms after delivery.^{1,2} ICP is a very rare condition, with a total incidence of 0.3%–0.5% in the general obstetric population. Although it is uncommon, it carries risks of fetal complications, such as spontaneous preterm labor, fetal distress, and intrauterine death, necessitating early detection and treatment for the best maternal and fetal outcome.^{1,3} Here, we describe a 30-year-old woman who presented with severe pruritus and was found to have recurrent ICP after she disclosed a history of identical symptoms that culminated in fetal death in her previous pregnancy.

Case Description

This is a case of a pregnant 30-year-old woman in her 18th week of pregnancy who presented with persistent itching, jaundice, and skin scratch marks. She had had a similar presentation in her previous pregnancy 4 years prior, which resulted in a miscarriage at 6 months. On physical examination, she had icteric sclera and skin scratch marks on her extremities and abdomen. Otherwise, her vital signs and other physical examinations were normal. Liver enzymes and bilirubin levels were increased (AST-71, ALT-57, ALP-337, with direct bilirubin 12.4 and total bilirubin 13.32 $\mu\text{mol/L}$), and serum bile acid levels were also significantly elevated (129 $\mu\text{mol/L}$ [0–10 normal range]). The CBC panel and coagulation parameters were otherwise normal, with no HBsAG, anti-HCV, or antimitochondrial antibodies, and ANA titer was normal. Blood film was also performed, revealing no hemoparasites, including *Plasmodium* spp. Later, she underwent abdominal ultrasonography and magnetic resonance cholangiopancreatography (MRCP), neither of which revealed any abnormalities. Eventually, following a presumed diagnosis of ICP, the patient was started on ursodeoxycholic acid (UDCA) at 13 mg per kg per day to be taken

orally, which was continued till delivery. After 1 month on medication, her pruritus had improved significantly, and her bile acid levels had returned to normal within 2 months of treatment. The delivery was also uneventful, with an outcome of a live female neonate.

Discussion

ICP is one cause of pruritus that presents without a rash and has more severe consequences than other causes of itching.⁴ It is thought to have genetic, hormonal, and environmental causes, though the exact causes are as yet unknown. Mutations in phospholipid and bile salt-export pumps found in the liver and placenta of ICP patients are believed to be the major contributing factors.⁵ Those women who have liver disease, family history of ICP, multiple gestations, or have undergone in vitro fertilization have a higher risk than others.⁶ The diagnosis of ICP is based on the presence of pruritus of cholestasis, elevated fasting serum bile acids $>10 \mu\text{mol/L}$, elevated serum transaminases, spontaneous relief of signs and symptoms within 2–3 weeks after delivery, and absence of other diseases that cause pruritus and jaundice.²

The ICP-management approach focuses on relieving maternal symptoms and ensuring a successful pregnancy outcome. UDCA, which displaces hydrophobic bile acids to protect hepatocytic membranes and stimulate transplacental elimination of bile acids from the fetus, can alleviate the maternal symptoms.⁷ Closer monitoring of the fetus and possibly earlier delivery at 37 weeks have been found to reduce fetal morbidity and mortality in ICP-complicated pregnancies.⁸

Conclusion

The appropriate diagnosis combined with UDCA treatment and vigilant obstetric follow-up had significantly reduced maternal symptoms and may have prevented a possible intrauterine death in our patient.

Data Sharing

Supporting data for the current case report are available from the corresponding author on reasonable request.

Ethics Approval and Informed Consent

This case report was approved by the Research Ethics Review Committee of the School of Medicine, Addis Ababa University. Prior to data collection, written informed consent was acquired from the patient after the studies had been well explained, and the patient also provided written informed consent for the case details to be published.

Disclosure

The authors report no conflicts of interest in this work.

References

1. Puhl T, Beuers U. Intrahepatic cholestasis of pregnancy. *Orphanet J Rare Dis*. 2007;2:1–6.
2. Knox TA, Olans LB. Liver disease in pregnancy. *N Engl J Med*. 1996;335(8):569–576. doi:10.1056/NEJM199608223350807
3. Kenyon A, Tribe RM, Nelson-Piercy C, et al. Pruritus in pregnancy: a study of anatomical distribution and prevalence in relation to the development of obstetric cholestasis. *Obstet Med*. 2010;3(1):25–29. doi:10.1258/om.2010.090055
4. Lee RH, Greenberg M, Metz TD, Pettker CM. Society for maternal-fetal medicine consult series# 53: intrahepatic cholestasis of pregnancy: replaces consult# 13, April 2011. *Am J Obstet Gynecol*. 2021;224(2):B2–B9.
5. Begley NR, Beltran AF. Intrahepatic cholestasis in pregnancy: increased surveillance and the role of bile acids in a patient with history of fetal demise. *Obstet Gynecol Cases Rev*. 2021;8:212.
6. Kamimura K, Abe H, Kamimura N, et al. Successful management of severe intrahepatic cholestasis of pregnancy: report of a first Japanese case. *BMC Gastroenterol*. 2014;14(1):1–5.
7. Zhang L, Liu X-H, Qi H-B, et al. Ursodeoxycholic acid and S-adenosylmethionine in the treatment of intrahepatic cholestasis of pregnancy: a multi-centered randomized controlled trial. *Eur Rev Med Pharmacol Sci*. 2015;19(19):3770–3776.
8. Glantz A, Reilly S-J, Benthin L, et al. Intrahepatic cholestasis of pregnancy: amelioration of pruritus by UDCA is associated with decreased progesterone disulphates in urine. *Hepatology*. 2008;47(2):544–551. doi:10.1002/hep.21987

International Medical Case Reports Journal

Dovepress

Publish your work in this journal

The International Medical Case Reports Journal is an international, peer-reviewed open-access journal publishing original case reports from all medical specialties. Previously unpublished medical posters are also accepted relating to any area of clinical or preclinical science. Submissions should not normally exceed 2,000 words or 4 published pages including figures, diagrams and references. The manuscript management system is completely online and includes a very quick and fair peer-review system, which is all easy to use. Visit <http://www.dovepress.com/testimonials.php> to read real quotes from published authors.

Submit your manuscript here: <https://www.dovepress.com/international-medical-case-reports-journal-journal>