Case Report

Congenital tuberculosis causing hydrops fetalis: A case report and review of literature

Eshita Bansal, Shuchi Mehra, Kapil Bhalla

Department of Paediatrics, Pt. B.D. Sharma PGIMS, Rohtak, Haryana, India

Abstract

Tuberculosis (TB) is an infectious disease of which congenital TB is a rare form even in TB-endemic countries such as India. There are very few case reports of the same in the literature. Though the incidence rate of congenital TB is low, mortality rates are very high. Here, we report a case of a 2-day-old neonate who presented to Pediatrics Accident and Emergency with complaints of fast breathing and swelling all over the body. The baby had swelling all over the body and subcutaneous edema suggestive of hydrops fetalis. She was investigated and subsequently diagnosed to have congenital TB for which appropriate treatment was started. The baby is still on regular follow-up with no active complaints.

Keywords: Communicable diseases, congenital tuberculosis, hydrops fetalis

Introduction

Congenital tuberculosis (TB) is a rare manifestation of a very common infection in India. It may be found in children born to mothers infected with TB, acquired during intrauterine life or during passage through the birth canal. The term perinatal TB collectively refers to true congenital and neonatal forms of the disease. Although India is the highest TB burden country in the world, cases of congenital TB have hardly been reported and remain largely untreated, leading to the death of infants.

Hydrops fetalis is a condition in the fetus characterized by an abnormal collection of fluid with at least two of the following:

- Subcutaneous edema (more than 5 mm).
- Ascites
- Pleural effusion
- Pericardial effusion.

Address for correspondence: Dr. Eshita Bansal, House No.-494, Sector-14, Rohtak – 124 001, Haryana, India.
E-mail: eshibansal@hotmail.com

Received: 12-06-2023 **Revised:** 13-07-2023
Accepted: 15-08-2023 **Published:** 21-12-2023

Access this article online

Quick Response Code:

Website: http://journals.lww.com/JFMPC

DOI: 10.4103/jfmpc.jfmpc_959_23

It may further be classified as immune or nonimmune hydrops fetalis (NIHF). Various causes of NIHF include cardiovascular, hematologic, chromosomal, lymphatic dysplasia, inborn errors of metabolism, and infections.

Case Report

A 2-day-old female neonate was brought to the emergency with a complaint of fast breathing. The baby was born to a primigravida mother at 35 weeks of gestation (preterm) via normal vaginal delivery with a birthweight of 1.8 kilograms, appropriate for gestational age. A history of delayed crying was present. No adverse perinatal events such as history of birth asphyxia, leaking pervagina, pregnancy-induced hypertension, or gestational diabetes mellitus were reported. The baby was not administered the Bacillus Calmette–Guèrin (BCG) vaccine. She also showed generalized swelling and subcutaneous edema.

On examination, the heart rate was 140 bpm and respiratory rate was 66/min. The baby's abdomen was distended, and ascites were present. The spleen and liver could not be palpated. On bilateral chest examination, crepitations were present. Appropriate treatment was initiated. The mother was being administered

How to cite this article: Bansal E, Mehra S, Bhalla K. Congenital tuberculosis causing hydrops fetalis: A case report and review of literature. J Family Med Prim Care 2023;12:3412-4.

antitubercular treatment (ATT) for active intestinal TB since the third trimester and had been on ventilatory support since delivery.

Following this, various investigative procedures were performed. Blood investigations showed Hb of 12.5 g, TLC of 5000/uL, polymorphs of 67%, and platelet count of 80000/uL. Cultures of blood and urine were negative. Serology for Toxoplasmosis, Other agents, Rubella, Cytomegalovirus, Herpes simplex (TORCH) infections (T. gondii, Herpes simplex virus (HSV), rubella, cytomegalovirus (CMV), parvovirus, and HIV) was done and found to be negative. Rh incompatibility and ABO blood groups incompatibility as causes of immune hydrops fetalis were ruled out. Other causes of hydrops (nonimmune) such as cardiovascular, hemolytic, chromosomal, and inborn errors of metabolism were investigated and found to be absent.

The chest X-ray showed miliary shadows in bilateral lung fields [Figure 1]. An ultrasonography was performed, which indicated the presence of thickenings in the mesentery and hyper-echoic calcified foci in both the lobes of the liver. Abdominal Computerised Tomography (CT) revealed peritoneal thickenings and confirmed calcified foci in both liver (segment 7) and spleen (mid-pole) [Figure 2]. Serial gastric aspirate was obtained for two days to detect the presence of Acid-fast bacilli (AFB), the results of which were positive on both days. CSF analysis did not reveal any signs of tubercular meningitis. The sepsis screen was also negative.

Based on the radiological findings, a diagnosis of congenital TB was made. The baby was started on category 1 Anti-Tuberculosis Therapy (ATT)—isoniazid (H), rifampicin (R), pyrazinamide (P), and streptomycin (S) (2 months of isoniazid + rifampicin + pyrazinamide + streptomycin (2HRZS) + 4HR). For hydrops, furosemide was administered. She was taken home after 14 days. She was brought for a follow-up one and half months later. The baby was gaining weight as per standard norms (20–30 gm/day) and was accepting and tolerating ATT well.

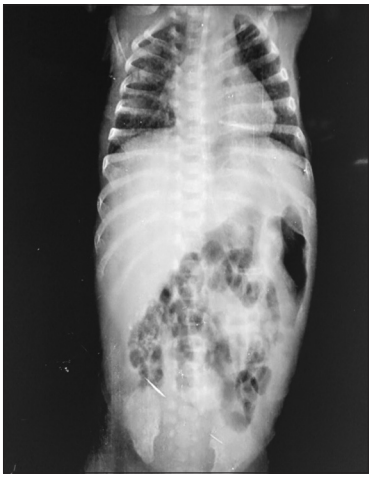

Figure 1: Chest and abdomen radiograph

Discussion

TB is a major public health problem in India. The country has the largest number of TB cases in the world—over a quarter of the global TB and multidrug-resistant TB (MDR-TB) burden.

The average prevalence of all forms of TB in India is estimated to be 5.05 per thousand, the prevalence of smear-positive cases is 2.27 per thousand, and the average annual incidence of smear-positive cases is 84 per 1,00,000 annually.

TB is generally caused by droplet inhalation. TB in the mother increases the risk of neonatal mortality and morbidity such as prematurity, low birth weight, acute fetal distress, and perinatal death.

In newborns, the infection may be congenital (in utero) or acquired after birth. Congenital TB may be transmitted by the hematogenous route via the placenta or by ingestion or aspiration of amniotic fluid. Placental transmission occurs via the umbilical vein and may result in hepatic granuloma along with the involvement of periportal lymph nodes. Another possibility is that the dormant bacilli in pulmonary circulation cause primary TB after birth due to increased oxygenation and circulation. Aspiration or ingestion or infected amniotic fluid can result in multiple primary foci in the lungs or Gastro-Intestinal Tract (GIT).

Congenital TB is particularly challenging to diagnose due to the similarity of symptoms in congenital TB and other neonatal infections. Also, the absence of AFB in any samples collected makes it difficult to draw conclusions.

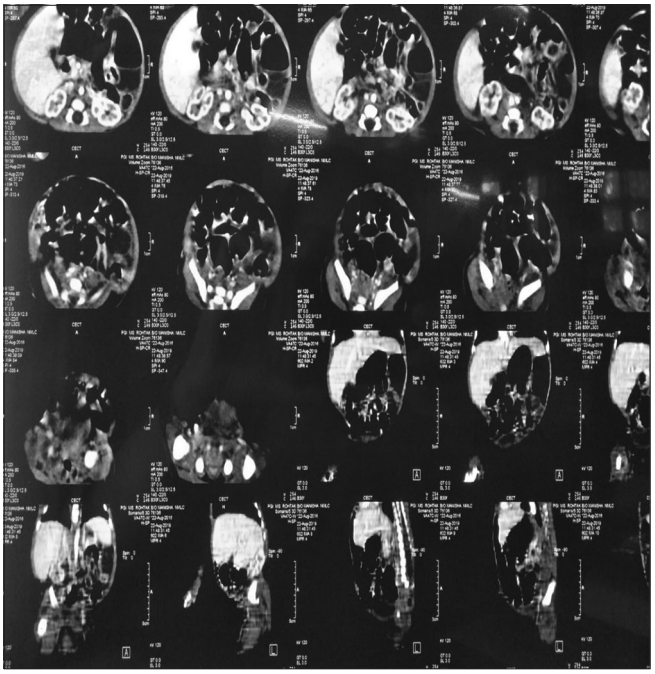

Figure 2: CT of the abdomen

Criteria were formulated by Cantwell *et al.* to diagnose congenital TB[1-3]—a proven tuberculous lesion plus one of the following:

- Lesions in the first week of life
- Primary hepatic complex or caseating hepatic granuloma
- TB infection of the placenta or maternal genital tract
- Exclusion of the possibility of postnatal transmission.

The clinical manifestations of congenital TB, mainly include hepatosplenomegaly (76%) followed by respiratory distress (72%), fever (48%), abdominal distension, and lethargy.[1,4]

The literature was reviewed for cases of congenital TB presenting with or leading to hydrops fetalis. Similar cases have been reported, by J Cheon *et al.*,[5] Y. H. Chung *et al.*,[6] and S. Chen[7], which highlight the need for well-defined guidelines for perinatal diagnosis of TB. Y.H.

In the case report by J Cheon *et al.*,[5] a 37-year-old findings of fetal ascites and pleural effor fetal hydrops and preterm labor at 29 weeks of gestation with no maternal serological evidence of recent toxoplasmosis, CMV, rubella, or HSV infection. A live girl was delivered at 29 weeks, weighing 1.5 kg, with an Apgar score of 6 at five minutes. The baby developed increasing consolidation of the right lung, and Mycobacterium TB was cultured in pleural effusion. Despite intensive support, the baby died at 11 days of age.

Chung *et al.*[6] reported a 37-year-old primiparity woman in her 26th week of gestation with findings of fetal ascites and pleural effusion. Despite fetal treatment of ascites and pleural effusion, the patient underwent an emergency cesarean section in her 29th week of gestation, due to a non-reassuring fetal heart rate. The infant died on day 18 and was diagnosed with congenital TB. The mother had no symptoms related to lung or extrapulmonary organ invasion caused by TB. The mother was diagnosed with latent TB during the postpartum period. As a result of this case, the authors recommended that physicians suspect latent TB as one of the causes of (NIHF and include maternal infection of TB in the evaluation of NIHF, particularly in countries with a high prevalence of TB.

In general, NIHF occurs due to the accumulation of interstitial fluid that fails to return to the venous system. This may further be due to cardiac failure, impaired venous return, obstruction to normal lymphatic flow, increased capillary permeability, or decreased oncotic pressure.

However, the relationship between congenital TB and hydrops fetalis cannot yet be fully understood as it has not been studied well.

Conclusion

Here, we have reported a case of congenital TB with hydrops fetalis, presenting on the 2nd day of life, who was saved and thrived due to timely diagnosis and intervention. Although congenital and postnatally acquired neonatal TB is difficult to distinguish clinically, it has great epidemiological significance. Perinatal TB cases may be largely underreported due to unawareness leading to the death of infants before a diagnosis can be reached. The mortality rate of congenital TB is nearly 50%. Improvement in poorly defined guidelines for prenatal diagnosis of congenital TB and heightened awareness to consider diagnosis could aid in attaining better outcomes. Children born to mothers infected with TB must be screened for any signs of transmission. In such cases, congenital TB must be kept as an important differential, especially in endemic countries. Early diagnosis and management are crucial to tackle these rates.

Acknowledgements

All authors approved the final version of the manuscript and are accountable for all aspects related to the case report.

The authors would like to express gratitude to the faculty and staff of the Department of Paediatrics, Pt. B.D. Sharma PGIMS, Rohtak, for all the support and suggestions given.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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