

## Case Reports

### **Congenital Tuberculosis: An Underdiagnosed Entity**

**M. Verma**  
**J. Chhatwal**  
**Y.K. Sarin**  
**V.P. Singh**  
**T. Singh.**  
**R. Aneja**

Congenital tuberculosis is an infrequently encountered condition although tuberculosis *per se* is continuing to be a major public health problem in our country. Among the few cases reported from our country, five were proven cases while one was of probable congenital tuberculosis (1-6). The age at time of diagnosis as well as the clinical picture of each of these cases was different and in all but two cases the outcome was fatal. Considering this, one needs to have a high index of suspicion to make the diagnosis. The clinical presentation of congenital tuberculosis is variable (7) and a clinician may overlook the diagnosis unless one specifically looks for some definitive clues on history, examination and investigations. We report here two cases of congenital tuberculosis with unusual clinical presentations.

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*From the Department of Pediatrics and Pediatric Surgery, Christian Medical College and Hospital Ludhiana, Punjab.*

*Reprint requests: Prof. M. Verma, Professor and Head, Department of Pediatrics, Christian Medical College and Hospital, Ludhiana 141 008, Punjab.*

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*Case I:* A 54-days-old male infant was admitted to the Department of Pediatrics with history of persistent respiratory tract infection since 21 days of age. He was a pre-term baby born at 34 weeks of gestation weighing 1.3 kg at birth. The antenatal period had been uneventful and there was no history to suggest intrauterine infections. The infant had received multiple antibiotics prior to being hospitalized. On examination, he was a pale, malnourished infant weighing 3 kg. He was dyspneic, had subcostal and inter-costal recession and coarse crepitations all over the chest. Liver and spleen were enlarged, 4 cm and 1.5 cm respectively below the subcostal margin. Other systems were essentially normal. Investigations revealed: hemoglobin-9.8g/dl, TLC 6,400/cu mm with 86% polymorphs and 4% band cells. His CSF was clear and blood culture was sterile. The chest radiograph showed confluent bronchopneumonia. As he had been unresponsive to prior antibiotic therapy, he was also investigated for tuberculosis. His Mantoux test and gastric aspirate smears for acid fast bacilli (AFB) were negative. Laryngeal swab grew *pseudomonas*. He was treated with cefotaxime, gentamicin and chloromycetin. A repeat X-ray done on the 16th hospital day showed some clearing of pneumonia but also showed a pneumatocele suggestive of staphylococcal infection (Fig. 1). Methicillin and rifampicin were added and the baby was discharged after 32 days of hospital stay. A repeat chest X-ray done after 40 days of treatment showed persistence of pneumatocele and infiltrates. As there was no significant radiological improvement

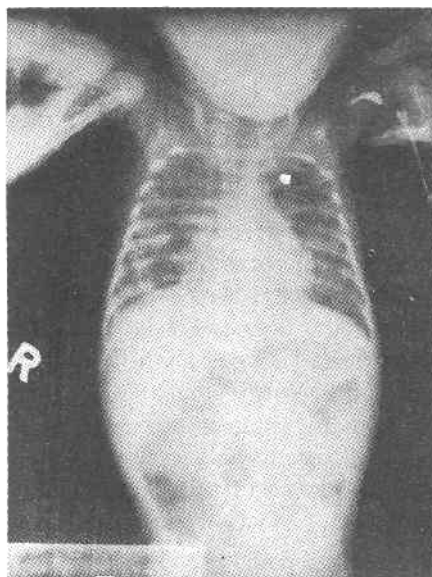


Fig. 1. X-ray chest of Case I.

and hepatosplenomegaly persisted, a possibility of tuberculosis was again considered and a liver biopsy was performed which revealed tuberculous granuloma with typical Langhans giant cells. Subsequently, an ultrasound revealed nodes at porta hepatis in addition to hepatosplenomegaly.

This indicated that what were being considered pneumatoceles were actually tubercular cavities. The patient's presentation and the course of illness had been modified as he had received amino-glycosides (gentamicin) which do affect mycobacterium tuberculosis. He was started on two drug antituberculous regimen (rifampicin and isoniazid). Later on, after he had gained weight and muscle mass, streptomycin was added. The mother was also investigated and diagnosed to have tuberculosis and started on antitubercular treatment.

*Case II:* A 3-month-old male infant was admitted in the Pediatric Surgery unit with history of vomiting, obstipation and abdominal distension of 1 week duration. On examination, he was malnourished, dehydrated and had mild jaundice. Systemic examination revealed a distended abdomen with visible peristalsis. There was no hepatosplenomegaly. He was diagnosed to have intestinal obstruction and operated. Operative findings showed dense adhesions between intestinal loops, other organs and peritoneum. The histopathology of bowel and lymph nodes was consistent with a tuberculous infection. In the meantime, the past treatment papers were brought by the parents which showed that the baby had been diagnosed to have tuberculosis at 2 months of age. He presented with symptoms dating back to 1 month of age and at that time had hepatosplenomegaly. The X-ray chest done was normal: however, Mantoux test was strongly positive and mother had been recently diagnosed to have tuberculosis. On the basis of above, the infant had been started on INH, rifampicin and pyrazinamide which he had received for 1 month when he presented to us. The jaundice was probably due to hepatotoxicity of anti tubercular drugs. We withheld these drugs till his liver functions returned to normal and then he was started on streptomycin, INH and rifampicin with steroids. The infant had a hectic surgical course but made a good recovery and at present is on oral feeds, passing stools and gaining weight.

### Discussion

An awareness regarding the varied manifestations of congenital tuberculosis would help in an early diagnosis. The criteria for making the diagnosis of congenital tuberculosis were set out in 1935 by Beitzki (8). These were that the diagnosis

must be bacteriologically proven and that the disease must be present within the first few days of life. In cases presenting later in life, there must be a primary complex in the liver or a possibility of extrauterine infection should be excluded by separating the baby from the mother at birth and segregation from any other possible source of infection. Case I unequivocally qualifies the diagnosis of congenital tuberculosis as a primary complex w as histopathologically proved to be present in the liver. The clinical picture and the course of the disease of this patient were probably affected by the aminoglycosides and rifampicin given as a part of anti pyogenic therapy. Our Case II fails on count of exclusion of post uterine infection by contact, but tuberculin test was strongly positive at 1 month of age. As the usual interval for hypersensitivity to tuberculosis (as denoted by skin test) after exposure is about 6 weeks (9), we believe that infection in Case II was most probably intrauterine. Beitzke's criteria, laid about 60 years ago, are considered too rigid now. Some authors feel that distinguishing congenital from early neonatally acquired tuberculosis is a matter of primarily epidemiological importance and the modes of presentation, treatment and immediate prognosis do not strikingly differ from the cases of congenital tuberculosis designated using Beitzke's criteria(10,11). Recently, it has been suggested that detection of identical phage types of *Myobacterium tuberculosis* in the mother and her infant would support the diagnosis of congenital tuberculosis (12).

Miller recognizes three different types of cases that should be labelled as congenital tuberculosis (7). The first are infants with a primary focus in the liver and a mass of lymph nodes at the porta hepatis. Few of these patients may have primary complex of the lung also. The second group do not have a primary lesion in the liver but have large numbers

of tuberculous foci scattered throughout both the lung fields and caseation in hilar and mediastinal lymph nodes. The mode of infection in the first group is transplacental, whereas in that of second group is infection through aspiration of amniotic fluid, material from the genital tract, or mouth to mouth breathing by an infective adult at child's birth. Besides these two groups, Miller also includes a third group which is a very rare presentation. These infants have abdominal primary tuberculosis, as seen in our second case, and the mode of transmission of infection is oropharyngeal taking place at birth or shortly afterwards (7).

The clinical picture of congenital tuberculosis can be variable. The age at the time of presentation has been noted to be few hours after birth (5) to beyond neonatal period. The signs and symptoms noted in various cases are shown in *Table 7*(10). Other rare manifestations of the disease include meningitis, otitis media, masto-itis, and skin lesions. The diagnosis is based on positive smear and/or culture results obtained from gastric washings, liver biopsy, lymph node biopsy, spinal fluid, ear discharge, endotracheal

*TABLE 1-Signs and Symptoms Noted in Congenital Tuberculosis*

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Main findings
- Respiratory distress
- Fever
- Hepatic and/or splenic enlargement
- Poor feeding
- Lethargy and/or irritability
- Lymphaderopathy
Other findings
- Abdominal distension
- Failure to thrive
- Ear discharge
- Skin lesions
- Jaundice
- Seizures

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aspirate, or bone marrow biopsy(12). The tuberculin test is only rarely positive.

Insufficient data is available to recommend any regimen of drugs, but it is believed that at least 2 and preferably 3 drugs should be administered. Hagemen *et al* (10) feel that streptomycin needs to be omitted, but in view of severe hyper-bilirubinemia seen in Case II, we preferred to use streptomycin in conjunction with isoniazid and rifampicin.

Because of the efficacy of modern antituberculous drugs, early diagnosis has become critical to affect a favorable outcome. Such early diagnosis is possible only if the disease is considered in the differential diagnosis. This becomes all the more relevant in our circumstances where a significant number of pregnant women may have active tuberculosis.

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