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Perinatal Tuberculosis with Spinal Involvement: a Case Report and Review of the Literature

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Summary

AkinyoolaAL, Adejuyigbe EA. Perinatal Tuberculosis with Spinal Involvement: a Case Report and Review of the Literature. *Nigerian Journal of Paediatrics* 2005; 32: 51. Perinatal tuberculosis is a rare condition. We present the case of a three-month old baby girl with perinatal tuberculosis which involved the thoracic spine. The diagnosis was based on the clinical features, positive tuberculin skin test and typical radiological findings. The patient has made a remarkable improvement on a combination of antituberculous drugs and spinal immobilization. This is the first reported case of perinatal tuberculosis with spinal involvement in Ile-Ife.

Key words: Tuberculosis, Perinatal, Spine.

Introduction

TUBERCULOSIS caused by *Mycobacterium tuberculosis* is one of the intractable diseases in the tropics. According to the World Health Organization, there are about 8-10 million new cases of tuberculosis per year in the world with approximately, three million deaths. The HIV pandemic has increased the prevalence of tuberculosis especially among women of reproductive age.[1] Presently, perinatal tuberculosis is very rare even in human immunodeficiency virus (HIV) endemic populations.[2] However, the incidence is likely to increase as tuberculosis continues to spread among women of reproductive age.[1] Only about 300 cases of congenital tuberculosis have been described so far[2] and most reported cases in the perinatal period are congenital. The diagnosis is usually made late by liver biopsy or at autopsy, except where there is a high index of suspicion.[2,3] A child may be infected before or during birth via the umbilical cord, aspiration of infected amniotic fluid and direct contact with tuberculous cervicitis or endometritis.[1,4,5] The liver, lungs and lymph nodes are the commonly affected organs.[1-4] Perinatal tuberculosis with involvement of the spine is very rare[2] and we are not aware of any reported case in our environment. Hence, this report of a three-month old girl seen at the Obafemi Awolowo University Teaching Hospitals Complex (OAUTHC), Ile-Ife.

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Case Report

Baby OS, a three-month old girl was referred to the Children Emergency Room (CHER) of the OAUTHC with a history of cough since birth, difficulty with breathing for four weeks and swelling on the back for one week. She was initially taken to a Comprehensive Health Centre where she was treated with antibiotics without relief. The cough and breathlessness had been intermittent while the back swelling had progressively increased in size since it was first noticed. There was associated fever and irritability at onset.

The mother received antenatal care and had normal spontaneous vaginal delivery with cephalic presentation at a private hospital. She denied any history of cough during and after pregnancy. However, the father had a chronic cough for which he had never sought medical attention. There was no history of cough in the siblings. The child was on exclusive breast-feeding but was yet to receive any immunization. Both parents were farmers.

Examination showed an ill female infant who weighed 4.5kg with a length of 48cm and an occipito-frontal circumference of 41cm. Her hair was fluffy and brownish in colour. She was dyspnoeic and tachypnoeic with a respiratory rate of 58 breaths/minute. She was afebrile with an axillary temperature of 36.5°C. Her heart rate was 150 beats per minute, and heart sounds I and II were normal. Fine crepitations were heard over the right hemithorax and she had a firm, non-tender hepatomegaly of six cm below the right costal margin. There was a kyphosis in the middle thoracic spine at D5 to D8 level with a left sided firm, tender paraspinal swelling extending

from D5 to D10 and measuring 6cm by 10cm. There was no neurological deficit in both upper and lower limbs. The laboratory investigations revealed a packed cell volume (PCV) of 38 percent, white blood count of 10.4 x 10^9/L with a differential of 23 percent neutrophils, 67 percent lymphocytes and 10 percent monocytes. Her platelet count was 386.0 x 10^9/L and the erythrocyte sedimentation rate was 82mm/hr (Westergren method). Analysis of her gastric aspirate for acid fast bacilli was negative as was retroviral (HIV) screening of the child and her parents. Chest radiographs showed clear lung fields, and kyphoscoliosis with destruction of the fifth to eighth

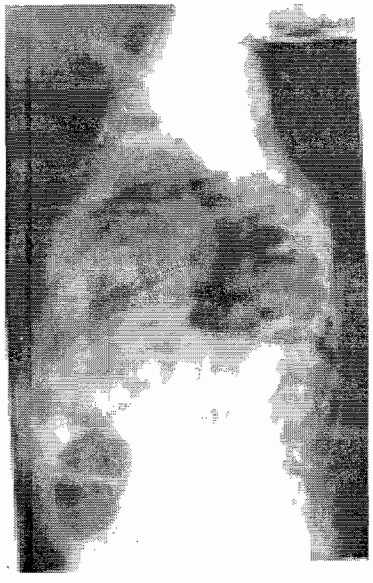

Fig 1: Plain radiograph (AP) of the child showing clear lung fields and destruction of D5 to D8 vertebral bodies

thoracic vertebral bodies (Figs. 1 & 2), while the tuberculin test was positive (13mm). Chest radiographs of both parents were normal but the father's tuberculin skin test was positive (17mm).

The child was commenced on a combination of intramuscular streptomycin (20mg/kg), oral rifampicin (20mg/kg), isoniazid (10mg/kg) and pyrazinamide (25mg) for the intensive phase, while rifampicin and isoniazid were continued thereafter. Thoracolumbar jacket made of scotch cast was applied to stabilize the spine, while her father was referred to the respiratory clinic of OAUTHC where he was treated for pulmonary tuberculosis. Our patient was discharged home two weeks after the commencement of the anti-tuberculous drugs with marked improvement. She attended the paediatric orthopaedic and infectious out-patient clinics for follow-up. At the first follow-up clinic, six weeks after commencement of antituberculous drugs, the signs of improvement noticed included reduction in the severity and frequency of coughing, disappearance of dyspnoea and rapid increase in weight, which was now 5.7kg. Six months after commencing treatment, the kyphosis had virtually disappeared and the child was thriving well. This was the last time the patient was seen and all efforts to trace her whereabouts have proven abortive.

Discussion

Tuberculosis during pregnancy may result in infection of the placenta or the maternal genital tract.[4,5] Such infection is then transmitted to the foetus by haematogenous spread from the placenta to the umbilical veins or by the aspiration or ingestion of amniotic fluid contaminated by placental or genital infections.[1,4] Haematogenous spread leads to the formation of one or more primary complexes in the liver or lungs. Aspiration or ingestion of infected amniotic fluid results in primary complex formation in the lungs or gastro-intestinal tract, respectively.

Co-infection with HIV, an important contributing factor in the recent rise in tuberculosis, increases the likelihood of extrapulmonary tuberculosis.[6] However, our patient and her parents were HIV negative. Involvement of the spine is one of the common extra-pulmonary manifestations of childhood tuberculosis.[7] It accounted for about 61 percent of cases of extra-pulmonary tuberculosis in northern Nigeria, most commonly affecting the thoracic spine.[8] We are however, not aware of any report of perinatal

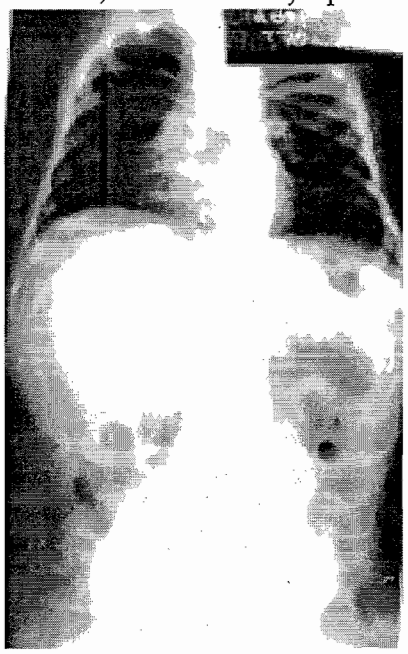

Fig 2: Plain radiograph of the chest and abdomen (lateral view) showing kyphosis and destruction of D5 to D8 vertebral bodies

or congenital tuberculosis with spinal involvement in our environment. Grover *et al*, reported an 18-day-old baby with congenital spinal tuberculosis in India; the case was diagnosed by imaging studies.[2] Airede working in Jos, reported a case of a premature baby who had the classical features associated with congenital tuberculosis and presented a diagnostic challenge;[3] diagnosis was confirmed by liver biopsy

and the child responded well to isoniazid and rifampicin. Manji *et al*,[9] reported the first case of presumed congenital tuberculosis from Tanzania.

There is need for a high index of suspicion especially in environments where maternal HIV and tuberculosis are highly prevalent. In a review of the English literature by Hageman *et al*,[10] the median age of presentation was 24 days and the presenting symptoms and signs were often non-specific. The mothers had sub-clinical tuberculosis that was detected only after the disease in the infants was diagnosed, reinforcing the importance of a thorough evaluation of mothers of infants with suspected congenital tuberculosis. Beitzke in 1935, established the standardized criteria for distinguishing congenital from postnatally acquired tuberculosis.[1] These criteria are (a) proven tuberculous lesions in the infant and (b) one of the following lesions in the first few days of life: (i) a primary hepatic complex, and (ii) the exclusion of postnatal transmission by the separation of the infant at birth from the mother and other sources of infection. These criteria are very difficult to apply in current practice and have been met in only a few cases.[1] The exclusion of postnatal transmission relies on a thorough investigation of contacts including the infant's hospital attendants. An open surgical procedure or autopsy is necessary to demonstrate a primary hepatic complex and regional lymph node involvement. Hepatic complexes are seldom demonstrated because few autopsies are performed; in addition, liver biopsy are now increasingly being performed percutaneously.[1] The presence of caseating hepatic granulomas permits the differentiation of congenital tuberculosis from a postnatally acquired infection on the basis of liver biopsy findings alone.[10] Abdominal ultra-sonography and other imaging studies are now increasingly used to demonstrate hepatic granulomas.[2]

Because of the rigidity and lack of easy clinical application of Beitzke's criteria, Cantwell *et al* in 1994, proposed revised criteria for the diagnosis of congenital tuberculosis. The criteria are (a) a proven tuberculous infection of the infant and (b) at least, one of the following:

(i) lesions in the first week of life;

(ii) a primary hepatic complex or caseating hepatic granuloma;

(iii) tuberculous infection of the placenta or maternal genital tract; and

(iv) exclusion of postnatal transmission by a thorough investigation of contacts. The symptoms of cough, breathlessness, fever and irritability in our patient were present at birth suggesting perinatal tuberculosis. Although there was hepatomegaly, we could not confirm hepatic granuloma because liver biopsy and ultrasonography were not done.

Infection of the placenta or the maternal genital tract is necessary for congenital transmission.[1,4,5,10] In Hageman's series, all the 14 mothers of infants with congenital tuberculosis had genital tuberculosis. It has been difficult convincing the mother of our patient to agree to gynaecological and microbiological evaluation.

The treatment of postnatally acquired tuberculosis is similar to that of congenital tuberculosis.[4,5] Neonates should receive isoniazid (10 to 15mg per Kg body weight per day), rifampicin (10 to 20mg per kg per day), pyrazinamide (20 to 30mg per kg per day) and streptomycin (20 to 30mg per kg per day) for the first 2 months, followed by isoniazid and rifampicin for four to 10 months depending on the severity of the disease.[4,5] Our patient was still on isoniazid and rifampicin with satisfactory improvement when she was last seen.

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