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Case Report

Infantile tremor syndrome

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ABSTRACT

Infantile tremor syndrome (ITS) is a clinical entity characterized by tremors, skin pigmentation, anemia, developmental and mental retardation. It is most commonly found in Indian subcontinent and in children who are exclusively on breast feeding of vegetarian mother. Amongst all the etiological theories, nutritional theory is the most accepted one. Management of ITS is largely empirical and includes therapy of nutritional deficiency, anemia & tremors apart from appropriate care. Treatment of nutritional deficiency includes vitamin B12, multivitamins, folic acid, iron, calcium, zinc, magnesium and high protein diet. In this article, three cases of ITS with classical picture have been reported.

Keywords: ITS, Tremor, Anemia

INTRODUCTION

Infantile tremor syndrome (ITS) is a clinical condition of acute or gradual onset of mental and psychomotor changes, pigmentary disturbances of hair and skin, pallor, and tremor in malnourished children aged between 5 months and 3 years & most commonly found in Indian subcontinent.[1,2] Though it is a well-known clinical entity, the etiology, pathogenesis and management of this condition remains obscure.[3] Etiologic theories for ITS including malnutrition, vitamin B12 deficiency, magnesium deficiency and infections have been studied, but the theory of nutritional deficiency is most accepted till yet among all. Many researchers have tried to correlate the symptoms with certain trace of element deficiency states without satisfactory results.[4]

CASE REPORT

Case report 1

A two years old female child, born normally at full term, fully immunized and on exclusive breast feeding presented with history of sudden onset of involuntary movements (tremors) in right leg and rapidly involving whole body, head & tongue without loss of consciousness. Her weight was 6.9 kg and height was 67 cm. She had pallor, dull expressionless face, hypopigmented sparse hair & delayed developmental milestones. On systemic examination, respiratory system revealed bilateral occasional ronchi. On central nervous system (CNS) examination, she was found to have tremors & irritability and tremors disappeared while sleeping. Per Abdominal and cardiovascular (CVS) examination showed no abnormality.

Investigations were suggestive of severe macrocytic hypochromic anaemia (Hb 6.4 gm%), RBC 2.48 mill/cmm, Mean corpuscular volume (MCV) 82 fl, Packed cell volume (PCV) 22.6%, Mean corpuscular hemoglobin (MCH) 26 pg, Mean corpuscular hemoglobin concentration (MCHC) 28.4%, Red blood cell distribution width (RDW) 26% with peripheral smear showing hypochromic macrocytic picture. Her serum calcium value was low (9.4 mg/dl), while Serum B12, Random Blood Sugar (RBS), serum TSH (Thyroid

stimulating hormone), Cerebrospinal fluid (CSF), urine and stool examination were normal. Chest x-ray, Magnetic Resonance Imaging (MRI) brain and ultrasonography of abdomen were also normal. Based on above mentioned findings, patient was diagnosed to have "Infantile Tremor syndrome". Initially patient was fed with Ryle's tube feeding and blood transfusion was done. Child was started with vitamin A, vitamin C, Folic acid, vitamin B12, zinc, thiamine, pyridoxine, proteins, Phenobarbitone and other supportive treatment. Child showed gradual improvement. Tremors were reduced and patient was switched over to spoon feeding.

Case report 2

A twelve months old male child, born normally at full term, exclusively on breastfeeding & bottle feeding presented with history of tremors in left upper limb and rapidly progressed to whole body & head. Child's weight was 5.7 kg and height was 65 cm. On examination, child had pallor, hypopigmented sparse hair, knuckle pigmentation & delayed developmental history of milestones. CNS examination showed tremors and respiratory system examination revealed bilateral ronchi on auscultation. Cardiovascular and abdominal examinations were normal. Investigations revealed anaemia (Hb 10.6 gm%), total RBC 3.66 mill/cmm, MCV 92.62 fL, MCH 28.96 pg, MCHC 31.27%, RDW 23.60% with peripheral smear showing mixed variety picture (hypochromic-microcytic-macrocytic). CSF examination was normal. On admission, child was treated with Phenobarbitone, multivitamin syrup, vitamin A, vitamin C, Folic acid, vitamin B12, zinc and other supportive treatment. Child showed improvement after 10 days.

Case report 3

An 18 months old male child fully immunized having past history of Enteric fever and treated at other hospital previously presented with history of tremors in upper limbs and rapidly progressing to whole body (Figure 1). His weight was 8.2 kg and height was 70 cm. On examination child had knuckle pigmentation and mild developmental delay. Systemic examinations including respiratory system, cardiovascular system and abdominal system showed no abnormality. Central nervous system examination showed coarse tremors. Investigation revealed Hb 9.3 gm%, RBC 3.4 mill/cmm, MCV 110 fl, PCV 30.8%, MCH 27.03 pg, MCHC 30.19 % with peripheral smear suggestive of macrocytic hypochromic anemia picture. His serum calcium was 9.8 mg/dl and vitamin B12 level was 50.78 pg/ml. Urine routine, chest x-ray, tuberculosis, measles, HIV work up, CSF examination & MRI of brain showed no abnormality. Child was treated with folic acid, vitamin B12, vitamin A, other multivitamins & minerals along with Phenobarbitone & supportive treatment. Tremors decreased gradually and patient was discharged after 2 weeks.

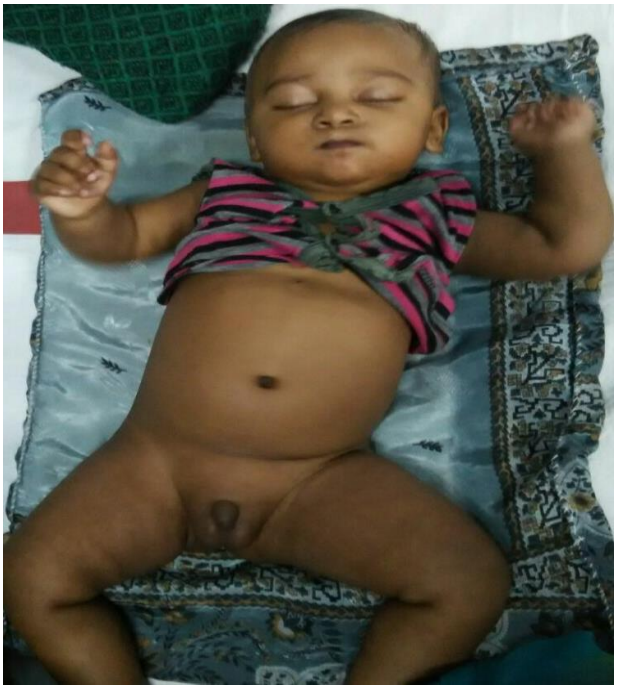

Figure 1: Hyperpigmentation in case report 3.

DISCUSSION

Infantile tremor syndrome is characterized by tremors, skin pigmentation, anaemia, developmental & mental retardation.[5] It is most commonly found in children belonging to poor socioeconomic status and who are on exclusive breast feeding of vegetarian mother, which showed similarity with our cases. Some studies reported that boys are more commonly affected than girls, while few studied showed equal gender distribution in occurrence of ITS.[4,6]

These three cases were presented with classical picture of ITS and were comparable to other cases described in literature. They were having tremor of acute onset, prominent in distal limbs and involving head & disappeared during sleep. Tremors are generally self-limiting and attributed to structural and functional changes of extrapyramidal system.[7,8]

Infantile tremor syndrome cases always present with various vitamins deficiency and varying degree of anemia.[4,9] Similarly these three cases were also found anemia with variable peripheral smear picture. In two cases peripheral smear was suggestive of hypochromic macrocytic anemia, while in one case it showed hypochromic-microcytic-macrocytic anemia.

Many theories have been studied for etiology of ITS, but still it remains obscure. Etiologic possibility of malnutrition was present in our all three cases. Management of ITS being largely empirical and it includes treatment of anemia and nutritional deficiency. Treatment of nutritional deficiency includes vitamin B12, multivitamins, folic acid, iron, calcium, zinc, magnesium

and high protein diet.[10] All our cases received Vitamin B12, iron, folic acid, multivitamin syrup, vitamin A, vitamin C, Zinc apart from appropriated care. Only one child required blood transfusion due to severe anemia. For the management of tremors, many drugs have been tried including Phenobarbitone, Chlorpromazine, Carbamazepine, Propranolol.[4,8] All our cases were treated with Phenobarbitione and favourable response was obtained.

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