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A Cumulative Approach on Rare Case of Infantile Tremor Syndrome

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Infantile Tremor Syndrome (ITS) is categorically determined as a self-limiting clinical presentation which is characterized by anaemia, tremors, pigmentary skin disease, muscular hypotonia, and mental development regression. The presenting tremors are coarse in nature which are either decreased or vanished during sleep and takes 4-6 weeks duration for complete resolution following its natural course. The aetiology for ITS has been hypothesized for various factors like metabolic, infectious, nutritional but remained inconclusive for effective statement. But the precipitation is on the developing deficiency of Vitamin B12 but it remained controversial in terms of different factors. As the aetiology remained undefined, the line of treatment for children with ITS has been considered as per the treatment for undernourished child which included nutritional management

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with external supplementation of trace elements like iron, magnesium, calcium, vitamin B12 along with other such multivitamins. The administration of tremors is most commonly managed with the administration of propranolol or phenytoin, phenobarbitone, and carbamazepine. An 8 months old male child was brought by parents with complaints of fever since 8 days, decreased appetite and vomiting since 3 days. The course of hospital followed by physiotherapy which helped in regaining the normal functional activity of the baby.

Keywords: Infantile tremor syndrome; ITS; tremors; undernutrition; tremors; deficiency; vitamin B12; physiotherapy.

1. INTRODUCTION

Infantile tremor syndrome (ITS) is a rare clinical disorder characterized by coarse tremors, anaemia and regression of motor and mental milestones. Most common in children of around 1 year of age [1]. The neurological consequences are reversible if diagnosed and treated at an early stage. ITS is categorically determined as a self-limiting clinical presentation which is characterized by anaemia, tremors, pigmentary skin disease, muscular hypotonia, and mental development regression. The presenting tremors are coarse in nature which are either decreased or vanished during sleep and takes 4-6 weeks duration for complete resolution following its natural course [2]. The aetiology for ITS has been hypothesized for various factors like metabolic, infectious, nutritional but remained inconclusive for effective statement. But the precipitation is on the developing deficiency of Vitamin B12 but it remained controversial in terms of different factors [3]. As the aetiology remained undefined, the line of treatment for children with ITS has been considered as per the treatment for undernourished child which included nutritional management with external supplementation of trace elements like iron, magnesium, calcium, vitamin B12 along with other such multivitamins [4]. The administration of tremors is most commonly managed with the administration of propranolol or phenytoin, phenobarbitone, and carbamazepine.

2. CASE REPORT

An 8 months old male child brought by parents from Amravati with complaints of fever since 8 days, decreased appetite and vomiting since 3 days. The baby was febrile since 8 days with on/off episodes, intermittent type, moderate grade, relieved on medication, no aggravating factors, vomiting occurred with 1-2 episodes since 2 days which was whitish in colour, non-bilious, non-blood stained, non-foul smelling. Mother also gave history of not accepting feeds well and he was disinterested in surroundings. The child was then taken to a local practitioner, where he underwent investigations which showed Haemoglobin as 4.5, Total Leucocyte Count (TLC) as 3500/cu.mm, platelets as 4 lacs with Typhoid positive (S.typhi O antigen positive) and then patient was brought for further management.

Child was born of a non-consanguineous marriage following a full term normal vaginal delivery to a primigravida mother at term gestation with birth weight of 2.5 kg, no history of neonatal intensive care unit (NICU) stay. He was an exclusively breastfed child and mother was strictly vegetarian.

Development history was normal. There was no history of developmental delay or no evidence of neuro regression.

On admission patient had tachycardia with heart rate (HR) as 158 beats/min, respiratory rate (RR) as 32 breaths/min, SpO_2 was 98%, and the peripheral pulsations were well palpable.He was poorly nourished with weight of 6 kilograms which was less than expected for age. Patient had pallor, thin sparse hair, mild tremors (involuntary movements) noted in the extremities of upper limbs. He also had hyperpigmentation of palms and soles of bilateral upper and lower limbs (especially knuckles), restless, lethargic and not interested in surrounding stimulii.

2.1 Anthropometry was noted

Weight- 6 kg (less than the expected weight for the age)
Height- 65 cm (less than the expected height for the age)
Head circumference- 41cm (less than the expected head circumference for the age)
Abdominal girth- 37cm

Systemic examination was within normal limits except for palpatory findings showing liver being palpable 1 cm below the costal margin.

Table 1. Anthropometry

	Right	Left
Midarm	12cm	12cm
Midforearm	11cm	11cm
Midthigh	20cm	20cm
Midcalf	15cm	15cm

2.2 Course in the Hospital

Complete work up including full blood count, renal abdominal liver function tests were done suggestive of Haemoglobin as 6.6g/dl, TLC as 4700/cu.mm, platelets as 56,000, reticulocyte count as 0.3%, Para check as negative, HPLC was also sent. Liver and renal function test were normal. Serum LDH was 1095. Strict heart rate and urine output monitoring was done. Supporting the finding of hyperpigmentation of extremities, parents also reported the history of consumption of vegetarian diet, so suspected deficiency was of vitamin B12, and hence vitamin B12 levels were sent which were 159 (reduced) confirming Megaloblastic anaemia (vitamin B12 deficiency). Patient was started on supportive line of treatment. Ophthalmic examination revealed normal fundus with optic disc with normal anterior segment.

Fig. 1. Clinical Presentation of the child

Considering this history and examination findings, a diagnosis of Infantile Tremor Syndrome was considered. Child was then given a course of injection Vitamin B12 1000 micrograms daily for 4 days. The patient was prescribed physiotherapy in an attempt to avoid any further secondary complication because of restricted activities. The physiotherapy was done during the inpatient stay of the baby which included the exercises for maintaining the configuration of individual joint and group of muscles. The physiotherapeutic session lasted for 20-25 minutes each day for 1 week.

Table 2. Timeline

Date of Admission:	19/05/2021
Date of Discharge	26/05/2021
Date of Physiotherapeutic assessment	27/05/2021
Date of Physiotherapeutic intervention	27/05/2021
Total duration of hospital stay	07 days

3. RESULTS

With appropriate treatment child gradually showed improvement in activity, accepting feeds well. tremors gradually decreased and completely stopped after 1 week of starting of the treatment.

4. DISCUSSION

Infantile tremor syndrome is a rare clinical disorder. Incidence in India approximately accounts for 0.2 to 2 % of hospitalizations [5]. The incidence had been reducing over the years, which can attributed to improved nutritional status, living conditions, better weaning practices and awareness of the parents about this clinical condition.

It commonly presents between 6 months to 18 months of age as neuro regression, malnutrition and acute tremor.

Tremors are initially intermittent but may become continuous in few days following an acute infection or stress. Tremors are more in distal parts of limbs, head, face and tongue and they disappear during sleep. These tremors are due to structural and functional alteration of extra pyramidal system [6].

Neuro regression in infants has different causes in which vitamin B12 deficiency is one of the uncommon causes. Infantile vitamin B12 deficiency is commonly seen in poorly nourished infants(especially infants of strict vegetarian mothers). In British Medical Journal 2 cases both infants of 8 and 10 months of age were studied

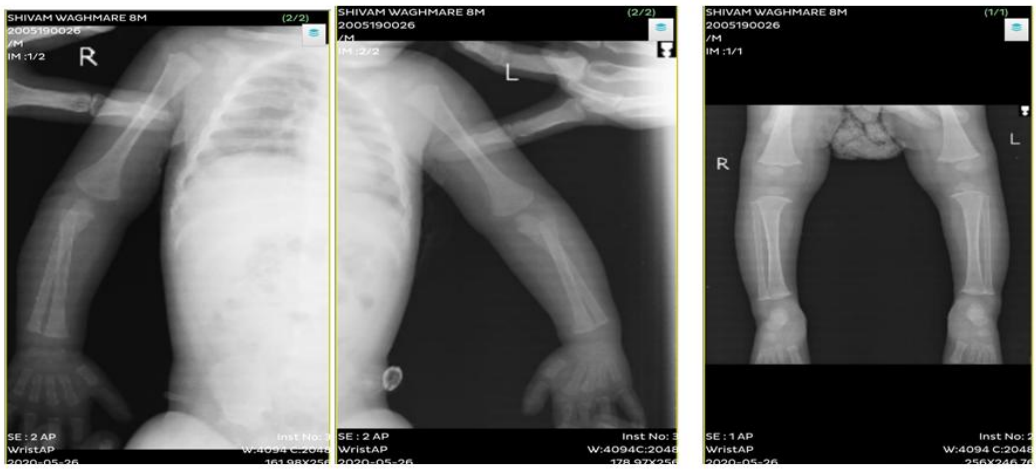

Fig. 2. X-ray images showing the reduced bone mass density

whose mothers are reported and documented to be vitamin B12 deficient. On admission, the patients were apathic, hypotonic and lethargic. Serum vitamin B12 levels were less than normal levels. Injection vitamin B12 was given and they had responded well to the treatment which was evidenced by improvement of clinical symptoms [7].

Prolonged breast feeding, pure vegetarian diet, iron, magnesium and zinc deficiency has been postulated in these poorly nourished infants in most of the cases. The neurological features can be accountable to the decreased levels of vitamin B12 and its transport protein Transcobalamin II in the CSF [8][9]. The CT and MRI findings are removed.

High doses of vitamin B12 may be required if serum B12 levels are low. Addition of multivitamins, vitamin C, iron, protein, zinc and magnesium supplements may also be useful [10]. Anemia can be micro or macro or normocytic in these cases [9].

5. CONCLUSION

Infantile Tremor Syndrome should be considered in a young child with neuro regression, tremor, developmental delay and malnutrition. The cumulative approach of medication and physiotherapy would be implicitly effective for the ITS.

CONSENT AND ETHICAL APPROVAL

As per international standard or university standard guideline Parental consent and ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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