

Infantile Tremor Syndrome - New observations

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Abstract

Infantile Tremor syndrome is a peculiar condition most commonly seen in Indian subcontinent. Although the number of cases with this syndrome reduced in recent years, finding of additional features made us to present this series. Total eleven cases were seen during Nov 2003 to Oct 2005 (3 year). Mean age of presentation was 11.2 months (range 6 to 18 months). The male to female ratio was 5: 6. There was no seasonal variation. Tremors, pigmentation, delayed milestones, pallor and hepatomegaly was present in all cases. Splenomegaly was present in 06 (54%) cases. LRTI was associated presenting feature in 09 (82%)cases and also CCF in 03 (27 %) cases. The striking feature noticed was presence of scurvy in all cases on x-ray imaging studies. All children received vitamin C, folic acid and vitamin B12 and antibiotics.

The mean duration of tremor phase was 23.2days with range 5 to 44 days. There was no mortality. Vitamin C and folic acid plays significant therapeutic role in ITS management.

Introduction

Infantile Tremor Syndrome (ITS) is a well known clinical entity. Although reported from outside India [1-3], seen more commonly in Indian subcontinent [4]. The clinical features of this syndrome are tremors, skin pigmentation, developmental and mental retardation and anaemia. But in recent years, in addition to the above characteristic features we noticed other findings [5]. Hence, we are presenting our observations on ITS in recent years.

Subjects and Methods

All children presenting with typical features of ITS, admitted in between November 2003 to October 2005 (3 years) in pediatric medical ward at Karnataka Institute of Medical Sciences, Hubli, Karnataka were analyzed.

A detailed history and physical examination was carried out. Investigations including complete haemogram, blood culture and sensitivity, urine microscopy stool microscopy.

CSF examination, chest x-ray, knee x-ray, Mantoux test and gastric lavage for AFB, HIV test were done for all children.

Results

The total number of cases seen during this period was 11. All children belong to age group between 6 months to 18 months with mean age 11.2 months and all of them belong to lower socioeconomic group. The male to female ratio

was 5:6. All children were exclusively breastfed till the time of admission except two in which in addition to breast feedings, small quantities of biscuits and rice were given. Out of 11 children, four each admitted during winter and rainy season and three in summer. Seventy three percent of (73%) them belong to severe malnutrition as per IAP classification.

Table I shows clinical features at presentation. In addition to characteristic features hepatomegaly was present in all cases and splenomegaly in 06 (54%) cases.

Three cases (27%) had hemoglobin (HB) less than 5gm/ dl and in remaining cases Hb varied between 5 to 10gm. Dimorphic anaemia was predominant picture (63%) on peripheral smear. Macrocytic anaemia was present in 4 (37) cases. There was no evidence of tuberculosis and HIV in any of our children. Urine and stool examination was normal and blood culture was sterile in all cases. CSF examination was within normal limits except in two cases where CSF protein was raised just above normal X-ray imaging studies revealed presence of scurvy changes in all children and pneumonic patches in seven cases (77%). C.T. scan head done in two cases was normal.

Table I Clinical features at presentation (n=11)

Sl. No.	Clinical features	No. of cases (%)
1. Tremors		
Localized	09	(82)
Generalized	02	(18)
2. Pigmentation		
Localized	07	(63)
Generalized	04	(37)
3. Pallor	11	(100)
4. Delayed milestones/ Mental retardation	11	(100)
5. Hepatomegaly	11	(100)
6. Splenomegaly	06	(54)
7. LRTI	09	(82)
8. C C F	03	(27)

All children received vitcofol (contains folic acid and Vitamin B12), and B12, Vitamin C and intravenous antibiotics. Three of these children required blood transfusion in addition to above medication. For control of tremors propranolol carbamazepine and phenobarbitone it one was used in 04, 01 and 01 cases respectively carbamazepine The total duration required for control of tremors in our cases ranged from 5 to 44 days with mean duration of 23.2 days. There was no mortality.

Discussion

We observed that girls outnumbered boys. In many series its been noticed that boys affected more commonly [6]. However, chaparwal et al in their study noticed equal sex distribution [7]. We also observed that there was no specific seasonal predominance in our cases where as earlier studies noticed seasonal variation [8-10] indicating probably other than viral etiology playing the role in causation of ITS. Apart from typical clinical features, we also noticed

presence of splenomegaly in more than in 50% of cases. Bajapai et al noticed splenomegaly only in 2 cases (out of 11). The probable reason for this high incidence in our cases could be due to simultaneous occurrence of LRTI.

The striking feature noticed in our cases was presence of scurvy changes on imaging studies. No other features of scurvy was present in any of our cases. The proposed mechanism explained elsewhere [5]. The mean duration taken for tremor control in our cases was 23.2 days with range from 5 to 44 days. Tandon et al reported the mean duration of tremors as 43.4 days with a range from 3 to 400 [12]. In other studies reported that mean being 50.5 days with range from 3-225 days [13]. Duration of tremor was reduced in our cases probably due to addition of vitamin C.

Majority of our children had LRTI at the time of presentation. Although it has been observed in earlier studies the presence of LRTI [14-15]. The exact reason for this high incidence of LRTI is not known but may be due to presence of less antioxidants due to deficiency of Vitamin C. In view of presence of scurvy and decreased duration of tremor phase with addition of vitamin c, we feel that folic acid and vitamin C plays a significant role in management of ITS.

In summary compared to the earlier reports, we in our children observed occurrence of ITS more in girls, no seasonal variation, Scurvy changes on imaging studies and decreased duration of tremor phase, in addition to this also seen higher incidence of LRTI and occurrence of splenomegaly.

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