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Carbamazepine Therapy for Infantile Tremor Syndrome

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Infantile tremor syndrome (ITS) is characterised by mental and psychomotor changes, pigmentary disturbance of hair and skin, pallor, tremors and subnormal

intelligence(1). Since the etiology of this disorder is largely obscure, the therapy has been empirical and supportive. Iron, magnesium, calcium, vitamin B<sub>6</sub> and B<sub>12</sub>, phenobarbitone and phenytoin have been used without consistent benefit(2,3). Kalra and Marwaha reported considerable improvement of symptoms in a trial of propranolol in eight patients(4).

Carbamazepine, a structural analogue of tricyclic antidepressants and phenothiazines, has been successfully used in treatment of various extrapyramidal movement disorders which include rheumatic chorea, chorea following head injury and torsion dystonia(5,6). We initially tried this drug on a child with ITS who was resistant to propranolol therapy. Dramatic disappearance of tremors in this child prompted us to use carbamazepine as a first line drug in four more children with ITS.

Case Reports

*Case 1:* An 11-month-old boy was admitted to Guru Tegh Bahadur Hospital, Delhi with the complaints of abnormal movements of limbs of four days duration following an episode of upper respiratory tract infection. The child showed global delay of developmental milestones. He was exclusively breast fed and was not weaned yet. On examination, weight was 7.5 kg and head circumference 45 cm. He was plump but pale with hair being depigmented, thin, silky and easily pluckable. Coarse tremors

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were noticed on all four limbs, in the perioral region and around eyes. These tremors disappeared during sleep. No focal neurological deficit was observed.

Investigations revealed a hemoglobin of 8 g/dl with dimorphic anemia. The cerebrospinal fluid analysis and radiographs of skull were normal. CT scan head showed mild cerebral atrophy. A clinical diagnosis of infantile tremor syndrome was made and the child was treated with propranolol (1.5 mg/kg/day) in two divided doses, besides supplementation with multivitamins (B<sub>1</sub>, B<sub>6</sub> and B<sub>12</sub>). There was no clinical response for ten days after starting therapy. Propranolol was discontinued and carbamazepine (20 mg/kg/day) in three divided doses was started. The tremors completely disappeared on the third day. The therapy was continued for eight weeks and then stopped. The child did not develop any untoward effects of carbamazepine therapy. The child has been followed up for 14 months. The tremors have not recurred though developmental delay persists.

*Cases 2-5:* Four patients of ITS were admitted in one year following the first patient. The clinical profile of these patients are shown in *Table I*. All were treated with carbamazepine (20-25 mg/kg/day) in three

divided doses. Three patients showed symptomatic improvement as evidenced by disappearance of tremors in 3 to 5 days duration. The fourth patient did not improve even after 14 days of therapy with carbamazepine, which was then discontinued. Patient was then treated with propranolol and the tremors disappeared on fifth day of this therapy. Carbamazepine therapy was continued for eight weeks in other three patients. None of them showed untoward effects of carbamazepine therapy. These patients have been followed up for 4 to 12 months after stopping therapy and tremors have not recurred.

### Discussion

Tremors in ITS are generally self limiting with the mean duration of tremors being 43.4 days (range 3-400 days)(2). Whether the duration of tremors has a direct correlation with the severity of residual defect is not clear. However, early control of tremors is extremely useful and shortens hospital stay. The cessation of tremors in four out of five patients within three to five days of starting carbamazepine therapy is very encouraging. In the first patient, carbamazepine therapy is very encouraging. In the first patient, carba-

**TABLE I—Clinical Profile of Patients Treated with Carbamazepine**

Case	Age (months)	Duration of tremors (days)	Previous developmental delay	Disappearance of tremors after therapy (days)
1	11	14	+	3
2	18	4	—	4
3	36	5	—	3
4	12	2	—	No improvement*
5	18	3	+	5

\* Tremors disappeared on propranolol.

mazepine was beneficial in controlling tremors resistant to even propranolol therapy.

The mode of action of carbamazepine is because of its structural similarity to phenothiazines(7). The drug blocks at the level of post-synaptic dopaminergic receptors. It also increases the activity of cholinergic system in cerebral hemispheres and corpus striatum which are antagonistic to dopaminergic system. Hence, it has been successfully used in disorders of extrapyramidal system including rheumatic chorea, chorea following head injury and torsion dytonia(5,6,8).

The clinical features such as tremors and dystonic posture in some infants in ITS suggests involvement of the extrapyramidal system. This prompted us to use carbamazepine therapy for this disorder. The drug is safe with few side effects when given in the recommended anticonvulsant dosages(7). Rash is seen in less than 5% patients and few develop reversible mild leucopenia. Blood dyscrasia and toxic hepatitis are extremely rare.

Carbamazepine was, therefore, effective in controlling tremors in patients with infantile tremor syndrome with no significant side effects. However, accepting the limitation of the study, we recommend further trials with carbamazepine in larger sample of patients before recommending it for routine use in patients with infantile tremor syndrome.

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## Amphotericin B in Visceral Leishmaniasis

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Visceral leishmaniasis has assumed epidemic proportions in north Bihar. Pri-

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