

MOVEMENT DISORDERS

OXCARBAZEPINE IN PAROXYSMAL DYSKINESIA

Four unrelated children (ages 8, 9, 16 and 17 years) with idiopathic paroxysmal kinesigenic dyskinesia responding to low dose oxcarbazepine are reported from the Department of Pediatric Neurology, Helen DeVos Children's Hospital, Grand Rapids, MI. Case 1, dyskinesias developed at 5 years of age and were induced by sudden purposeful movements such as standing up, running or jumping. Attacks consisted of abrupt cessation of activity, staring straight ahead with a fearful facial expression, followed by ballistic flailing movements of the arms and athetoid movements of the body. He fell to the ground but consciousness was maintained. Attacks lasted from 30 seconds to 2 minutes, and occurred up to 10 times a day. The general and neurologic examination, MRI brain, ictal EEG, and laboratory studies were all normal. Oxcarbazepine in a dose of 15 mg/kg/day controlled attacks without side effects, but parental withdrawal of medication after 6 months resulted in a recurrence that was reversed by restarting treatment. Case 2 developed a rash with oxcarbazepine; valproic acid was substituted and produced a 50% reduction in attacks. Other anticonvulsants tried, including levetiracetam, topiramate, and carbamazepine, were ineffective. Cases 3 and 4 also responded to oxcarbazepine. A trial of carbidopa/levodopa in case 4 was ineffective. Associated disorders were ADHD in case 1 and migraine in case 2, (Chillag KL, DeRoos ST. Oxcarbazepine use in paroxysmal kinesigenic dyskinesia: report of four patients. *Pediatr Neurol* April 2009;40:295-297). (Respond: Dr Chillag, 1300 Michigan Street NE, Suite 102, Grand Rapids, MI 49503. E-mail: kipp.chillag@devoschildrens.org).

COMMENT. Paroxysmal kinesigenic dyskinesia (PKD) is an involuntary movement disorder precipitated by purposeful movements or by startle, hyperventilation, stress, and photic stimulation. Movements are dystonic, choreic, athetoid, or ballistic, often in combination, unilateral or bilateral. Short attacks (few seconds to 1 minute) occur several times a day. They respond to antiepileptic drugs, especially oxcarbazepine. Other forms of paroxysmal dyskinesia are nonkinesigenic (induced by fatigue, alcohol, caffeine, or stress); hypnogenic (occurring during sleep); and exercise-induced. Duration is short or long-lasting, and etiology is idiopathic or secondary. An autosomal dominant genetic factor is involved in some familial cases, with evidence for a channelopathy. Many cases of paroxysmal hypnogenic dyskinesia are a form of frontal lobe epilepsy (Margari L et al. *Pediatr Neurol* 2005;32:229-235). A Pubmed search found 189 references, the latest by van Rootselaar AF et al. *Pract Neurol* 2009;9:102-109. The clinical evaluation of idiopathic PKD and diagnostic criteria are discussed in Bruno MK et al. *Neurology* 2004;63:2280-2287. In comparing familial and sporadic cases, sporadic cases were more frequently male, and infantile convulsions were more common in the familial kindreds.