

# Shuddering attacks

## A benign phenomenon in children

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### Abstract

**Question** A 2-year-old child was recently brought into my office for repeated episodes of neck stiffening and shivering movements of the shoulders and arms. The episodes last 4 to 5 seconds and occur more than 10 times per day, with no apparent pattern except increased frequency at mealtime. Although there has never been loss of consciousness, the parent was worried that these were seizures. The child was diagnosed by a neurologist as having shuddering attacks. Should I start antiepileptic medications for this child?

**Answer** Shuddering attacks are involuntary movements of the head and upper extremities that occur during normal activities and do not impair consciousness. Normal neurologic examination findings and normal electroencephalogram tracing will confirm that this child has shuddering attacks, a benign phenomenon that requires no further investigation or medical therapy. The condition is of unknown cause but is distinct from epilepsy and neither warrants nor responds to antiepileptic medications. Parents can be reassured that attacks will decrease in frequency and will spontaneously remit with age.

Shuddering attacks are brief episodes of shivering that can occur at high frequency, resemble seizures, and cause substantial concern among parents. It was first reported in 1976 as a harbinger of essential tremor in 6 infants and children with family history of tremor,<sup>1</sup> but subsequent reports failed to confirm this association.<sup>2,3</sup>

The incidence of shuddering attacks is thought to be very low, but misdiagnosis is common. In 2 tertiary hospitals in western Saudi Arabia, 12 children with shuddering attacks were identified during a 6-year period, none of whom was referred with the accurate diagnosis.<sup>3</sup> In 1 centre in Germany, 3 children were diagnosed within 1 year.<sup>4</sup> In Sydney, Australia, over 10 years, 21 of 666 children who underwent electroencephalogram (EEG) investigation for paroxysmal events were diagnosed with shuddering movements, accounting for 7% of 285 nonepileptic events.<sup>5</sup>

### Presentation

Shuddering attacks typically present as stiffening, tonic posturing, and rapid shivering movements of the head and upper extremities, at times involving the trunk.<sup>1,2,5,6</sup> Facial expression can change during an attack, including staring,<sup>2,4,6</sup> eye deviation, lip tightening, and teeth clenching.<sup>6</sup> Events typically last from a few to 15 seconds.<sup>2,5-7</sup> Frequency of attacks varies widely but can be as often as hundreds of times per day,<sup>1,8</sup> and episodes can occur in clusters of longer intervals.<sup>6</sup> Shuddering attacks differ from common shivering in their longer duration, greater frequency, and posturing of the arms.<sup>2</sup>

Children remain fully conscious throughout and after an episode, distinguishing shuddering attacks from seizures.<sup>2,7,9</sup> Attacks have paroxysmal onset during normal activities, such as when sitting in a chair or when

having a diaper change.<sup>2,6</sup> They occur spontaneously and are not elicited by voluntary movement,<sup>2</sup> although a 15-month-old boy had episodes associated with pressing Lego bricks together and had head movements when drying his hair.<sup>4</sup> Triggers of attacks include excitement,<sup>2,3</sup> eating,<sup>4,6</sup> and frustration with not being able to perform a task.<sup>3,6</sup> There is no report of shuddering attacks occurring during sleep.<sup>2</sup>

### Pathophysiology

The pathophysiology of shuddering attacks is unknown. The original report hypothesized that they are the immature brain's expression of essential tremor,<sup>1</sup> and the presentation of shuddering attacks can be similar to postural essential tremor.<sup>6</sup> However, a study of 39 children younger than 18 years of age with essential tremor found no personal or family history of shuddering attacks,<sup>10</sup> and other reports found no family history of essential tremor.<sup>3,6</sup>

One report suggested that shuddering attacks could be a variant of benign myoclonus of early infancy, which can present with similar movements and can also be triggered by eating and excitement.<sup>6</sup> However, this condition tends to have spasmic rather than tremulous movements and generally remits by 2 years of age.<sup>11</sup> Several patients had a history of imminent preterm delivery, suggesting a possible relationship with intrauterine stress.<sup>6</sup> Another suggested explanation is that shuddering attacks are a variant of motor tics, both exclusively occurring in wakefulness and precipitated by excitement or frustration.<sup>3</sup> However, only 3 children with shuddering attacks also had a history of tics,<sup>1</sup> and none had motor or vocal tics upon follow-up.<sup>3</sup>

Vitamin D deficiency has been suggested as a cause of benign shivers and tremors in newborns,<sup>12</sup> but only 1 report of shuddering attacks in children has included a

biochemical investigation and found a borderline level of vitamin D.<sup>7</sup> Other theories propose that shuddering attacks are seizures with deep-seated epileptiform discharges not detected on EEG, or that they are paroxysmal autonomic events similar to sneezing or yawning.<sup>9</sup>

### Diagnosis

Videotaping of episodes is helpful to distinguish shuddering attack movements from tonic or myoclonic seizures.<sup>3,4</sup> However, accurate diagnosis is best supported by documentation of normal EEG findings, with only muscle artifacts present during an attack.<sup>2,3,5,6</sup> Tracings show low-amplitude, high-frequency (8 to 10 cycles per second) motor movements, differing from the myoclonic or clonic movements in absence seizures, which are of higher amplitude and lower frequency.<sup>2</sup>

Findings of neurologic examination of children with shuddering attacks are found to be normal.<sup>2,3</sup> Growth parameters, including head circumference and developmental assessments, are generally normal in children with shuddering attacks,<sup>4</sup> although there are reports of diagnosis in children with mild developmental delay.<sup>2,6</sup> Blood tests are not needed, as blood glucose, electrolytes, and endocrine screening findings are normal.<sup>6,7</sup> Imaging of the brain is not recommended in children with this suspected diagnosis. Among 12 children diagnosed with shuddering attacks, 5 did computerized tomography scans and 3 did magnetic resonance imaging before specialist assessment, and all findings were normal.<sup>3</sup>

### Reassurance

Parental education and reassurance are all that is needed for this benign condition.<sup>3,4</sup> In 12 children with onset of shuddering attacks between 8 months and 2 years of age, all had complete remission by the age of 3 to 7 years.<sup>3</sup> In an 11-year-old girl with a 1-year history of shivering episodes, follow-up assessment at 8 months found no change in frequency, but the attacks were not adversely affecting the patient.<sup>2</sup> In a 3-year-old girl having several shuddering attacks per minute that interfered with feeding and dressing, a trial of 0.5 mg/kg per day of propranolol was effective for cessation of attacks within 1 week of therapy.<sup>8</sup> While  $\beta$ -adrenergic blockers are first-line therapy for essential tremor,<sup>13</sup> this is the only report of their use in shuddering attacks. After relapses by attempting to discontinue propranolol, the patient remained on half the initial dose indefinitely, with no report on long-term follow-up.<sup>8</sup>

Shuddering attacks are not associated with epilepsy later in life, although a report of a 2-year-old boy with attacks suggested development of status epilepticus.<sup>6</sup> Antiepileptic medications are not effective for shuddering attacks.<sup>14</sup> In a 21-month-old male infant with a family history of seizures in 2 first-degree relatives, a trial of antiepileptics (phenobarbital and ethosuximide) did not reduce the frequency of shuddering episodes.<sup>2</sup> Children who were prescribed antiepileptic therapy before a specialist evaluation were weaned off the medications with no effect on attack frequency.<sup>3,6</sup>

### Conclusion

Shuddering attacks are shivering movements of the head and upper extremities that typically last several seconds and can occur at high frequency. Normal neurologic examination findings and normal EEG tracing distinguish this condition from epileptic syndromes. Shuddering attacks spontaneously decline in frequency and eventually remit with age, warranting no pharmacotherapy. 🌸

#### Competing interests

None declared

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