

Transient infant movements (TIM): frequent infant non-pathological developmental motor phenomena

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VIDEO ONLINE

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ABSTRACT

Objective. Lombroso and Fejerman, in 1977, described non-epileptic movements in normal infants and named them “benign myoclonus of early infancy”, which were recently relabelled by Fernandez-Alvarez as “benign polymorphous movement disorder of infancy” (BPMDI). The focus of our study was to describe, categorize and point out the peculiar clinical representations of these heterogeneous phenomena through our video footage, particularly to those less experienced in paediatric neurology.

Methods. We included all infants with a video-EEG performed at our unit or a home video recording of “Fejerman-Lombroso”, “benign myoclonus of early infancy”, “shuddering attacks” or “paroxysmal non-epileptic movements”.

Results. Twenty-one children were selected. Age at onset ranged between two and 13 months, age at disappearance ranged between seven and 16 months, age at recording ranged between four and 16 months, and duration of the phenomena ranged between two weeks and 19 months. In total, 85% infants had normal neurodevelopment at onset and follow-up (mean follow-up: 31.47 months) and 15% presented with neuropsychological or neurosensory deficits. We distinguished four different patterns of movements: movement of the head in 50%, shuddering attacks in 30%, tonic brief contractions of the trunk and limbs in 10%, and elevation of the shoulders in 10%.

Significance. These motor phenomena do not affect neurological status and are not associated with developmental delay. Considering that clinical interpretation may be challenging, especially relative to epileptic seizures, we present an explanatory video of these characteristic events. We also propose a new definition that is simple to remember: “transient infant movements” (TIM).

Key words: benign myoclonus of early infancy (BMEI), benign polymorphous movement disorder of infancy (BPMDI), Fejerman-Lombroso, shuddering attacks, paroxysmal non-epileptic movements, transient infant movements (TIM)

Lombroso and Fejerman, in 1977, first described non-epileptic movements in normal infants as “benign myoclonus of early infancy” [1]. Dravet and colleagues in 1996 called them “benign non epileptic infantile spasms” and focused on the

differential diagnosis between this “benign myoclonus” and epileptic spasms of West syndrome [2]. Differences between the two manifestations were the normality of the EEG counterpart and the transient nature of the

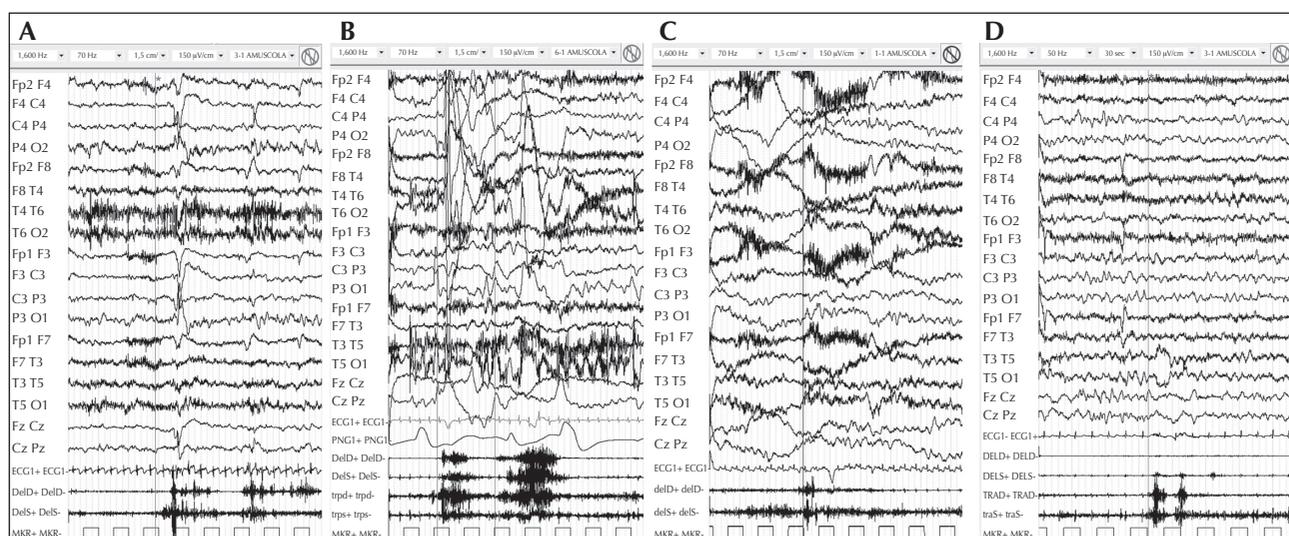
phenomena. In 1976, Vanasse, and colleagues described “shuddering attacks” through peculiar parents’ descriptions of the phenomena, assuming a possible correlation with essential tremor [3]. In 2008, Tibussek and colleagues also focused on shuddering attacks, pointing out the importance of reassuring parents to avoid extreme concerns [4]. Later, Pachatz and colleagues described two patterns: shuddering, referred to as axial paroxysmal motor manifestation involving mainly the trunk, and a newly described pattern of tonic limb contractions [5]. In 2009, Caraballo and colleagues recognized other phenomena within the same manifestations, such as myoclonic jerks, brief tonic contractions and atonia [6]. They also focused on searching for clinical characteristics that could be specific to these phenomena, such as occurrence only during wakefulness, or the differential diagnosis with other pathological conditions such as hyperekplexia, Sandifer syndrome and tonic reflex seizures of early infancy [6]. In 2012, Capovilla and colleagues focused on “head atonic attack” movement patterns, reporting cases of children who underwent drug therapy due to a misdiagnosis with epileptic seizure [7]. In 2018, Fernandez-Alvarez proposed the new definition “benign polymorphous movement disorder of infancy” (BPMDI), pointing out the clinical presentation of these events [8]. In the last year, Nagy and Hollody proposed videos for the attention of paediatric and child neurologists in order to facilitate the diagnostic process [9]. The aim of this study was to provide a description of a case series of paroxysmal non-epileptic movements

occurring in infants, in order to support paediatricians and parents in recognizing these manifestations and distinguishing them from other pathological movements, through the help of an explicative video recording. We also propose a new definition, with an acronym that is easy to remember.

Case series

From our database of video-EEG recordings, we selected all full-term infants with a diagnosis of “Fejerman-Lombroso”, “benign myoclonus of early infancy”, “shuddering attacks” or “paroxysmal non-epileptic movements” during the first year of life. The fundamental inclusion criterion, besides the onset of the manifestations within the first year of life (1-12 months), was presentation of the manifestations during a video-polygraphic/EEG, performed in our epilepsy monitoring unit. We also included three infants whose events were only recorded via home video, but the pathological disorders were ruled out based on the medical examinations. Poor-quality recordings were excluded.

The representative video sequence of the case series includes nine infants out of 21, for whom we obtained permission for publication by their parents. In *figure 1*, we show four examples of the polygraphic pattern of the different manifestations. In total, 21 infants (14 females and 7 males) were evaluated, all showing a normal EEG as expected.



■ **Figure 1.** (A) Head movement with contraction of the deltoid muscles. (B) Shuddering shown by the deltoids and trapezius muscles. (C) Tonic brief contraction of the trunk and limbs shown by the deltoid muscles. (D) Shoulder elevation shown by the trapezius muscle.

Head movements

Ten children among 21 (50%) showed predominantly head movements (see *video*; Infants 1 to 4).

We distinguished three types of head movements, all occurring during wakefulness:

- Five infants showed abrupt and repeated dropping of the head, while playing (two) or crying (one), or during a state of calmness (two). In three cases, these movements were associated with brief upper limb contractions or shoulder elevation, and in one case were associated with a facial grimace.
- In the other four children, we observed lateral lowering of the head. In three, this was repetitive, and in the other was a single movement with shoulder elevation.
- One infant showed a rapid jerk-like head movement, while crying.

The number of manifestations recorded were from a minimum of one to a maximum of 11, isolated or in brief clusters of 3-4 manifestations at a time. All single episodes lasted for one or two seconds, and a cluster no more than five minutes. The head movement was not isolated, but often accompanied by a concomitant movement of the upper limbs or shoulders, clearly visible on the EMG trace (deltoid, neck or trapezius muscles) and without impaired awareness. Age at onset varied from five to 13 months (average: eight months) and the manifestations disappeared at four months on average (from a minimum of 15 days to a maximum of 19 months). One child had neurosensory hearing dysfunction and another had language delay.

Shuddering attacks

Six children among 21 (30%) presented with shuddering attacks (see *video*; Infants 5 and 6). These consisted of brief shaking movements at high frequency during wakefulness and involved the whole body or a specific area of the body (as for the shuddering).

- Two children experienced a high frequency of shuddering during a state of calmness or upon photo stimulation, which was rapid and brief, lasting for a few seconds, involving particularly the upper limbs.
- One had shuddering involving the entire body which persisted for 40 seconds, while crying.
- Two children presented with a cluster of many consecutive shuddering episodes while playing, during excitation or in a state of rest.
- One infant presented with isolated shuddering attacks with a grimace triggered by excitation, while playing and eating.

Shuddering is characterized by rapid and arrhythmic muscle contractions, which occur asynchronously and gradually involving the head, shoulders, upper limbs, trunk and lower limbs, clearly visible on the EMG trace (deltoids and trapezius muscles) and without impaired awareness. All infants presented with brief manifestations, of one or two seconds in duration, with just one exception, Infant 6 in the video sequence, who had an episode lasting for 40 seconds. We recorded 1-11 manifestations. Age at onset varied from two to nine months (average: 5.3 months) and the manifestations lasted on average for 4.6 months (from a minimum of one month to a maximum of 10 months). In this group, one child presented with language delay and hyperactivity during our follow-up period.

Tonic brief contractions of the trunk and limbs

Three children among 21 (10%) presented with tonic brief contractions of the trunk and limbs (see *video*; Infants 7 and 8). These infants showed sustained contractions of the upper and lower limbs, isolated or in very brief clusters, several times a day, only during wakefulness and without impaired awareness. This phenomenon is also triggered by excitation and is characterized by rapid contractions clearly visible on the EMG trace (deltoid muscles). In our recordings, the infants presented with a minimum of seven and a maximum of 14 manifestations, predominantly isolated, which lasted from two to four seconds. Age at onset varied from four to nine months (average: seven months) and the manifestations disappeared at 7.3 months on average (from a minimum of one month to a maximum of 18 months). All children presented with normal neurodevelopment.

Elevation of the shoulders

Two children among 21 (10%) presented with shoulder elevation (see *video*; Infant 9). The episodes occurred isolated at the beginning, and then occurred more frequently and organized in brief clusters, consisting of abrupt elevation and abduction involving predominantly just one shoulder (visible on the EMG trace with deltoids and trapezius muscles) during a state of calmness or evoked by external noises, during wakefulness and without impaired awareness. We recorded three and nine manifestations in each child, respectively. In this group, the age at onset was seven months on average and the manifestations disappeared at one month in both infants. They had normal neurodevelopment during our follow-up period.

Discussion

Through our case series, we aimed to describe the different and multiple expressions of motor non-epileptic and non-pathological phenomena that occur in healthy infants during the first year of life. We underline the importance of video recordings in order to gather the peculiar clinical characteristics that could make their recognition easier, especially for those less experienced in paediatric neurology. According to our follow-up, we also confirmed that these phenomena have a self-limiting course with variable duration; although in all infants, they disappear within the second year of life. In doubtful cases, video/EEG with polygraphy recording is fundamental to exclude other pathological conditions. Considering the clinical features, we can summarize that these phenomena occur randomly during wakefulness, isolated or organized in brief clusters, affecting all areas of the body, but mainly the head and trunk, usually symmetrically. This phenomenon should be differentiated from epileptic spasms as it is not strictly stereotyped and does not occur periodically or correlate with sleep. Conversely, epileptic spasms usually occur during drowsiness or awakening and are stereotyped, reproducible and generally occur periodically and organized in clusters. These benign manifestations should also be differentiated from pathological myoclonus which is a sudden, brief, shock-like movement that may be repetitive or rhythmic, and lasts typically for no more than 200 milliseconds [10]. These episodes are usually longer (lasting for a few seconds), non-repetitive and each phenomenon appears to show different features associated with the movements (regarding duration, the muscles involved, and vehemence), both in the same child or between different children, even though the movement may be related to a specific pattern (head movement, shuddering, tonic contractions, shoulder elevation). The fact that they are often triggered by situations of excitation or frustration, particularly when infants interact with other people, is also a peculiar characteristic of these non-epileptic movements. Epileptic phenomena, in contrast, are generally not actuated by these particular situations. We suggest that these manifestations are referred to by the acronym “TIM” or “transient infant movements”, which is easy to remember and implies the age-dependent and self-limited aspects associated with these events, as well as the short duration of the phenomenon itself. With regards to what other authors have previously suggested, we believe that it would be less confusing not to use the term “movement disorders” as part of the definition, given the fact that these manifestations are solely paroxys-

mal movements, not pathological, and by implication, do not represent a disorder.

Conclusions

This paper describes and shows, through video sequences, some previously undocumented clinical characteristics of paroxysmal non-epileptic movements that appear during the first year of infancy in normal children, which may aid in better classifying the polymorphic nature of these non-pathological movements in infancy. We suggest that at the onset of the phenomena, simple video observation of these events should be adequate for paediatricians to hypothesize a benign aetiology. Later on, video-polygraphic/EEG recording is recommended in order to confirm the normality of cerebral activity and, when possible, record the events. We propose the name “transient infant movements” and its acronym “TIM” as a useful and practical term to describe these episodes. ■

Supplementary material.

Summary slides accompanying the manuscript are available at www.epilepticdisorders.com.

Disclosures.

None of the authors have any conflict of interest to declare.

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Legend for video sequence

Clinical features of TIM based on our case series. The video sequence shows the following in order:

- (1) A five-month-old boy showing head movements.
- (2) An eight-month-old girl presenting with head movements.
- (3) A 10-month-old girl experiencing head movements.
- (4) An eight-month-old girl presenting with head movements.
- (5) A seven-month-old girl experiencing shuddering.
- (6) A five-month-old boy showing prolonged shuddering.
- (7) An eight-month-old girl presenting with tonic brief contractions of the trunk and limbs.
- (8) A nine-month-old boy showing tonic brief contractions of the trunk and limbs.
- (9) An eight-month-old girl experiencing unilateral shoulder elevation.

Key words for video research on www.epilepticdisorders.com

Phenomenology: shuddering, head deviation, side-to-side axial movement

Localization: not applicable

Syndrome: non-epileptic paroxysmal disorder

Aetiology: non-epileptic paroxysmal disorder

TEST YOURSELF

- (1) When and why could it be useful to perform a video/EEG and polygraphy?
- (2) Why is it helpful to educate paediatricians in recognizing “TIM”?
- (3) Why is it useful to take time when faced with these manifestations?

Note: Reading the manuscript provides an answer to all questions. Correct answers may be accessed on the website, www.epilepticdisorders.com.
