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CASE REPORT

Infantile Masturbation in a Nigerian Child: A Case Report of a Rare Seizure Mimic

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Summary

Infantile masturbation is a benign, paroxysmal, non-epileptic condition with stereotypic stiffening of the body/limbs, copulatory-like thrusting/rocking of the pelvis and somatosensory symptoms. It usually occurs without direct genital stimulation thus, making it easily misdiagnosed as epilepsy. A 12-month-old girl presented with a one-month history of 'jerking' with no fever or altered consciousness. The event usually stopped with distraction. The frequency and duration of events worsened progressively. Epilepsy was suspected initially but a subsequent review of a video recording showed a conscious female child with intermittent rhythmic rocking/thrusting of the pelvis while lying prone, making epilepsy unlikely. Infantile masturbation was diagnosed and the parents were counselled on behavioural therapy (distractions). The frequency and duration of the events progressively declined until complete resolution by six years of age. The characteristics of the events remained similar overtime except for occasional tucking of her clothes into her vagina at age five years. This report is accompanied by a review of the relevant literature on infantile masturbation.

Keywords: Africa, Early childhood masturbation, Gratification behaviour, Gratification disorder, Infantile gratification, Seizure.

Introduction

Masturbation refers to self-stimulation of the genitals to derive pleasure. [1,2] Infantile masturbation (IM) is a variant of masturbation that occurs in infants and young children, usually without direct genital stimulation. [3–6] It is a benign, self-limiting, paroxysmal, non-epileptic condition with repetitive stereotypic stiffening of the body, copulatory-like movements such as thrusting of the pelvis and, sometimes, somatosensory symptoms like flushing, irregular breathing or grunting. [2,5,7]

Compared to the Western countries, fewer reports of IM have emanated from Africa, [3] including two case reports from Nigeria, [8,9] presumably due to misdiagnosis by clinicians. [6–8,10] Child health professionals need to be aware of IM as a rare, but possible, differential diagnosis of childhood epilepsy or movement disorders to avoid misdiagnosis. [7] Therefore, this is a report (following CARE guidelines [11]) of a case of a Nigerian child with 'jerking' movements, initially suspected to be epilepsy, but later diagnosed as IM.

Case Description

A 12-month-old female child was brought to the clinic by the parents, with a one-month history of

'jerking of the body, especially the hip.' There was no associated jaw-clenching, eyeball rolling, altered consciousness, fever or changes in behaviour or personality in-between events. She usually stopped jerking when called or distracted but sometimes resumed the activities afterwards; nevertheless, she did not exhibit negative reactions such as anger when stopped. Initially, the episodes were infrequent, each lasting about a minute and occurring mostly when she was about to sleep. Over the ensuing two months, the frequency and duration of the events increased, occurring all through the day with each event lasting 3-5 minutes and, sometimes, occurring during sleep with each lasting about a minute. It also became associated with deep breathing and restlessness. She had no prior head trauma or exposure to any toxic substance. She was the first-born to non-consanguineous parents (her father was a clergyman and her mother was a full-time home-maker). The pregnancy, labour, delivery and neonatal period were uneventful. Her developmental milestones had been normal till presentation. There was no past or family history of seizures, nor history suggestive of sexual abuse or parental neglect. Her physical examination, including the neurological system, was essentially normal.

Following the initial evaluation, the attending junior medical officer at the General Out-patient Paediatric Clinic suspected urinary tract infection (UTI) and prescribed oral antibiotics, although there were no urinary symptoms. However, the 'jerking' movement persisted, necessitating a re-presentation at the same clinic where epilepsy was suspected and electroencephalography (EEG) requested. Video-recordings of the episodes were also requested. A review of the mobile phone-made video-recording showed a child who repetitively rocked/thrust the waist up-and-downward while lying face-down on a bed but stopped when distracted (Figure 1 shows a still image captured from the video clips which are available at https://data.mendeley.com/datasets/t88jjxkkkp provided with parental permission). Infantile masturbation was suspected and the parents were educated on this disorder and they were counselled to consistently distract and discourage the child during the events. A month after, the parents happily reported that the events had become very infrequent, each lasting only a few seconds at that moment. The girl continued to have occasional brief episodes until six years of age (Timelines of her clinical evolution are shown in Figure 2). The characteristics of the events remained similar until five years of age when she occasionally tucked her clothes into her vagina (without direct hand stimulation) during the event. It was also observed at this moment that she aborted the events and showed remorse when reprimanded. Her growth and development, including psychosocial and academic performances, have been normal till the time of this report.

Figure 1: Child having repeated rhythmic thrusting of the pelvis during a 'jerking' episode while lying prone on the bed (Images used with permission of parents).

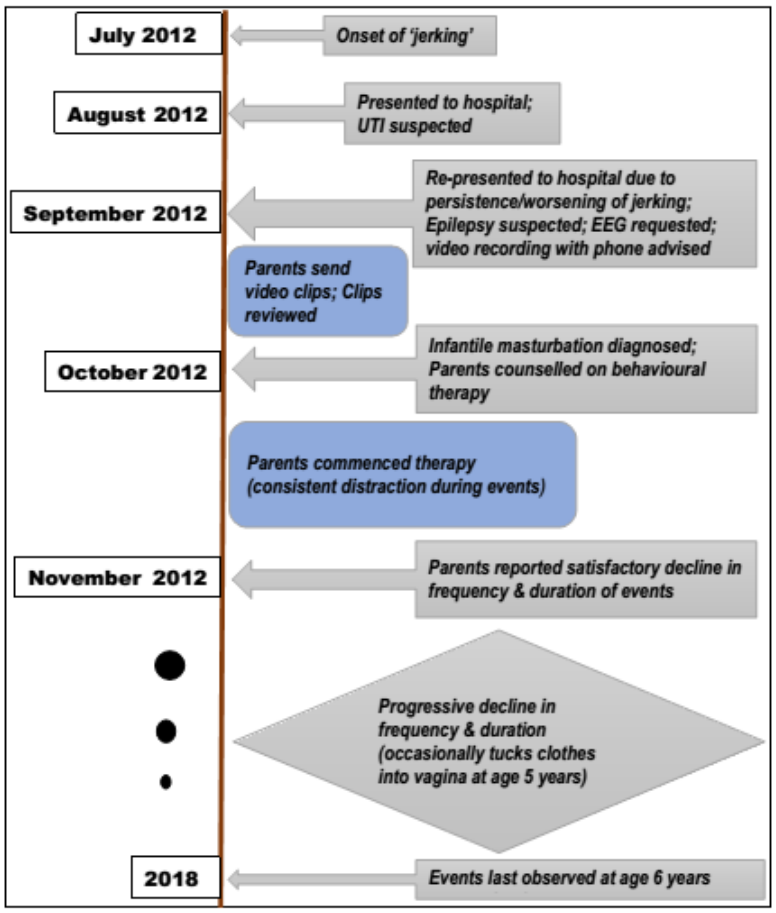

Figure 2: Timeline of management of infantile masturbation in a Nigerian child.

Ethical considerations

The authors obtained written informed consent from the parents to publish this report for research and academic purposes, including the public use of the images.

Discussion

With the aid of video-recording with a smartphone, the index case was diagnosed and successfully managed for infantile masturbation which was initially thought to be epilepsy. IM is frequently misdiagnosed as epilepsy because the affected children, unlike adults and older children, do not usually directly stimulate their genitals during the events. [7,12–14] It is a self-stimulatory behaviour, more appropriately termed "gratification disorder", "infantile gratification", "self-gratification", "gratification behaviour, or "early childhood masturbation." [6,10,13,15–18] To emphasise the associated motor elements, Nechay *et al* [19] and Phillips and Seshia, [17] proposed the use of the terms "benign idiopathic infantile dyskinesia" and "paroxysmal hyperkinetic motor syndrome of infancy" respectively.

The onset of IM is mostly in the first few years of life, as observed in the index case and previously-reported Nigerian cases. [8,9] There was a previous report of an in-utero masturbation event. [16] Otaigbe [8] reported a case of a 15-month-old girl with the onset of the symptoms and diagnosis at the 3rd and 15th month of life, respectively. Ibrahim and Raymond, [9] also reported an 18-month-old Nigerian girl who presented with a two-month history of abnormal movements subsequently diagnosed as IM after reviewing a video-recording. About half of the subjects in a British series developed IM before the first birthday. [14,15,19]

As observed in the index case, and previous reports from Nigeria, IM is commoner in females.

[2,19] In a retrospective case series of 31 British children described by Nechay *et al*, [19] females constituted 65% of the cohort. Female predominance has also been reported in other case series from North America, [6] Jordan [12] and Iran.[20] In contrast, Othman *et al*, [10] reported male predominance in a report of 11 cases from Sudan. The reason for this male predominance in Sudan is not immediately apparent; perhaps, it may reflect a cultural tendency towards better health-seeking behaviours by parents in North Africa for male children compared to female children. [21]

IM may present with various movement patterns including episodic stiffening of the body/pelvis, rhythmic rocking of the waist/pelvis (as in the index case), sometimes accompanied by dystonic posturing or scissoring of the lower limbs, [6,9,14] or rubbing of the thighs. [12] Somatosensory symptoms such as flushing, perspiration, vocalisation, grunting or irregular breathing may occur during the events; the index case exhibited 'deep breathing' and 'restlessness' during the events. [6,12,14] These somatosensory symptoms have been likened to orgasmic experiences in adults but they are essentially non-erotic in IM. [17,22] Some children may exhibit transient fixed or staring gaze ("watching of television in the air"). [8,23]

IM mostly occurs in the prone position because this position brings the genital area in contact with physical objects in the environment such as with the index case. [12,14,15,23] Other positions include supine or knee-chest; the events in the index case occurred only in the prone position. [12] Each episode may last up to 2-10 mins, or even hours occasionally, [10,12,17] with the frequency ranging from 2 to 20 times in a day. The events usually occur while awake or just before sleep, but may rarely occur during (the early phase of) sleep as occurred in the index case. This distinguishes it from benign sleep myoclonus of infancy which usually occurs during deep asleep. [2,12] Benign myoclonus of infancy which, although usually occurs while awake, is characterised by spasm-like myoclonic jerks of the limbs and neck was also excluded. The associated somatosensory symptoms in the index case and the tucking of clothes into her vagina further supports the diagnosis of IM and makes other non-epileptic paroxysmal disorders ('seizure mimics') of infancy like benign sleep myoclonus of infancy and benign myoclonus of infancy unlikely. [13,24]

IM is considered to be a normal psychosexual experience of early childhood. [1] However, it may be triggered by perineal irritation from vulvovaginitis, urethritis or pinworm infestation. [2,13,14] Children may respond to these perineal irritants by rubbing their thighs together; in the process, they accidentally 'discover' that such movements elicit pleasurable feelings which evolve into a habit with repetition. [2] Some children apply pressure on their suprapubic region to reinforce the pleasurable feelings. [6,15] Family stress/disharmony, boredom or tiredness may also trigger or aggravate the events as the child seeks an alternate source of affection. [5,10,12,14,15] In the index case, it was impossible to identify the likely trigger factor, although UTI was initially suspected, the diagnosis was not confirmed. It was also important to exclude sexual abuse in the index case because sexually abused children are more likely to have abnormal sexual behaviours compared with non-abused children. [1]

Children with IM are often subjected to needless investigations such as EEG, lumbar puncture, Computerized Tomographic scan, Magnetic Resonance Imaging, metabolic studies or gastrointestinal contrast studies, and antiepileptic therapy, with no resolution. [6,12,14,23,25] Although the diagnosis of IM is purely clinical, a high index of suspicion is required. [13,17,26] While there are currently no clear-cut clinical diagnostic criteria, the presence of certain features are regarded as highly suggestive of IM: (1) onset from the age of two months to four years, (2) stereotypic episodes of variable duration and frequency, (3) somatosensory symptoms like grunting, flushing, perspiration, (4) perineal pressure or stimulation with characteristic posturing of the lower limbs, (5) occurrence with intact sensorium but often just before sleeping, (6) cessation of events with distraction, (7) and normal physical and/or laboratory findings. [6,17] The diagnostic hallmark that has consistently distinguished IM from epilepsy is the cessation of the events with distraction (sometimes eliciting anger or temper tantrum, which did not occur in the index case). [4,7,12,23,27] The index case met all of the criteria for

diagnosis without the need for EEG. Nonetheless, EEG may occasionally be required to distinguish IM from epilepsies when history, examination and video recording of episodes are inconclusive. [18,28] Although EEG was requested in the index case, the parents were reluctant to do it because they were not convinced that their child had seizures, perhaps an expression of fear of stigmatisation commonly associated with epilepsy. The ubiquitous availability of mobile smartphones has made video-recordings greatly possible and more useful in the evaluation of movement disorders such as IM, further reducing the need for EEG. [24,27,29] Additionally, some authors have explored the potential roles of laboratory investigations such as serum oestradiol and urinary mucus in the diagnosis of IM but these are still largely inconclusive in the absence of adequately-powered prospective studies. [12,20]

An erroneous perception of IM as a 'spiritual problem', a feeling of stigmatisation and shame may discourage early presentation in the hospital, especially in Africa, where IM may be regarded as a taboo. [3,8,10,28] Otaigbe [8] and Negash and Bahiretibeb, [3] reported that parents were advised by neighbours to consider female genital mutilation as a cure for IM. This raised a possibility that IM could be a silent contributor to female genital mutilation in developing countries. [8,28]

Although benign, IM needs to be treated because of the associated parental anxiety, feelings of embarrassment/shame and cultural unacceptability. [1,3,8,28,30] IM has no specific therapy other than parental education (stressing its benignity) and behavioural therapy consisting of consistently distracting the child during the events or engaging the child in activities while awake to shift the child's attention to desirable activities such as playing with toys. [7,8,15,18,28] Frequent diaper change and the use of cotton underwear have also been used in its treatment. [20] The parents of the index case were educated using educational articles on IM and they were advised to consistently distract her during the events. Similar to previous reports, [8,9,15,19] the home-based interventions resulted in steady resolution of the symptoms over time.

Although most affected children stop manifesting symptoms by the age of one to three years, a few, as in the index case, may relapse occasionally as they grow beyond childhood. [7] IM is not associated with neuropsychological disorders subsequently [13,28] but some authors have reported a possible association with the development of an attention-deficit hyperactive disorder, [7] or Rett's syndrome [31] in later life. However, this is yet to be proven in appropriately-sized and -designed studies. Due to the significant possibility of bias and chance occurring with case reports, [11] it is impossible to completely exclude the fact that the episodes experienced by the index case may have been due to other causes or that the observed improvement may have occurred by chance. Yet, the observed pattern and course, including the response to distraction, support the diagnosis of IM. However, multicentre prospective cohort studies are required to reliably characterise the disorder, its management and long-term course, especially in African children.

Conclusion

IM may be misdiagnosed as epilepsy, with potential exposure to needless investigations and therapy, delayed diagnosis and parental anxiety. Video-recording is useful in reinforcing its diagnosis and it responds well to simple behavioural therapy with subsequent resolution of the events and parental anxieties.

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