

Original Research Article

Gratification phenomena in children: a report of nineteen children and review of the literature

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ABSTRACT

Background: Gratification phenomena (GP) are self-stimulatory behavior and also called as infantile masturbation. This phenomenon is especially seen in early childhood. GP is often misdiagnosed as epilepsy, dystonia, urinary tract infection or infantile colic resulting in numerous investigations and treatment because of variable behaviors during this event.

Methods: This study aimed to describe the features of GP, so that clinicians can better recognize this condition, to prevent unnecessary evaluations, and provide appropriate guidance to parents. This study is a prospective observational study, conducted in patients with GP between March 2019 and November 2020. GP was diagnosed on the basis of history, eyewitness account and video recording of the event

Results: During the study period, 19 children were diagnosed with GP. Out of 19, 17(89.4%) children were females. The mean age at onset was 17months (range 5-36months). The mean age at diagnosis was 20 months (range 8-47months). The mean frequency of events was 4/day (range 1-12/day). A typical GP was seen in 12 of 19 (63.15%) children. At the time of referral, 31% of children were diagnosed with epilepsy and 26% of children were diagnosed with infantile colic. During follow up period, GP was subsided within 4-12 months after the initial visit.

Conclusions: A GP, otherwise called infantile masturbation, is an important consideration in the differential diagnosis of epilepsy and other paroxysmal events in early childhood. A detailed history and video recording of events often prevents unnecessary investigations and treatments.

Keywords: Epilepsy, Gratification phenomena, Infantile masturbation, Paroxysmal events, Video recording

INTRODUCTION

Gratification phenomena (GP) in children are self stimulatory behaviors that tend to become a habit. Gratification phenomenon (disorder) also known as benign idiopathic infantile dyskinesia or infantile masturbation and is a form of non epileptic paroxysmal movement disorder.¹ It occurs in children between 3months to 3years old, but sometimes it can occur in school going children also.^{1,2}

It is a benign event and a normal behavioral variant seen in children.³ The typical features include lower limbs scissoring posture and repetitive pelvic movements with

rubbing the thighs together and rocking the genital region against the bed or other objects.⁴⁻⁶ The paroxysmal movements are typically associated with grunting, facial flushing, and irregular breathing, which are all followed by sweating and exhaustion.

The infant may not appear completely responsive with distant gaze, but the behavior can be interrupted by distraction. It is often misdiagnosed as seizures, dystonia, urinary tract infection or infantile colic resulting in numerous investigations and treatment.^{2,7} A proper history, eyewitness statement, and video recording of the event help in making the diagnosis. Pediatricians are in general aware of this phenomenon, but are perhaps less

aware of the spectrum of different patterns of this masturbatory activity.

In this clinical observation, we report a series of 19 children diagnosed with GP. This study aimed to describe the features of GP so that clinicians can better recognize this condition, to prevent unnecessary evaluations, and provide appropriate guidance to parents.

METHODS

This study was a prospective observational study, conducted at a tertiary hospital of South India during a period of 20 months (from March 2019 to November 2020). This study was approved by the institutional ethical committee. Written informed consent was obtained from parents of enrolled children.

Data for all the patients such as age, gender, birth history, developmental history, detailed history about events and follow-up were recorded. The diagnosis was based on accurate history and video recordings taken during event by parents.

The majority of children did not require further work up. Only some children underwent variable tests include electroencephalogram (EEG), Neuroimaging and blood investigations. Children are followed by direct clinic visits and telephonic interview for 6-15 months. The data were presented as number and percentages.

RESULTS

During the study period, total 19 children were diagnosed with GP. From these subjects, 17 of 19(89.4%) children were females. Neurological and developmental evaluations were age appropriate in all children. Family history was negative for similar events.

The most common reason for referral was for possible epileptic seizures in 6(31.57%) children. Other children were diagnosed with infantile colic (5), urinary tract infection (2) before attending our center. Six children are visited our center first time without any prior evaluation (Table 1).

Age at presentation was 8months to 47months with a mean age of 20months. Age of onset was 5months to 36months with a mean age of 17months. Nine of 19(47.36%) had onset less than 12months of age. The frequency of events varied from 1 to 12/day (mean of 4/day, median 7/day). The mean length of events was 4minutes (range 30seconds to 30min).

Different patterns of GP were seen in children. Typical GP was seen in 12(63.15%) children. Behaviors during events included scissoring in 12, leg twisting in 11, leg rising in 8, grunting noises in 6, rocking in 6, and sweating in 4. One child each displayed cyanosis, lip smacking and appeared frightened. All the children stopped their events

with distraction. None had events in sleep. Thirteen children were evaluated with routine blood investigations, urine examination, and ultrasound abdomen before referral. At the time of referral five infants (26.3%) were diagnosed as infant colic and on probiotics, antispasmodic drugs. Six (31.57%) patients were diagnosed with epilepsy and were on antiseizure drugs. Only two children had abnormal urine examination and treated with antibiotics. EEG was performed in 12 children and there were no epileptiform discharges. Six children had MRI brain prior to our assessment with no reported abnormalities.

Table 1: Clinical features and investigations in children with gratification phenomena.

Variable	Value
Sex	
Female	17 (89.4%)
Male	2 (10.6%)
Age at presentation	8-47 months (Mean 20 months)
Age at onset	5-36 months (Mean 17 months)
Age of onset <1year	9 (47.36%)
Patterns of gratification	
Typical	12 (63.15%)
Scissoring	12 (63.15%)
Leg twisting	11 (57.89%)
Leg rising	8 (42.1%)
Grunting	6 (31.57%)
Rocking	6 (31.57%)
Sweating	4 (21.05%)
Frequency	1-12/day (Mean 4/day)
EEG done	12 (63.15%)
MRI Brain done	6 (31.57%)
Diagnosis prior to refer	
Epilepsy	6 (31.57%)
Infant colic	5 (26.31%)
UTI	2 (10.05%)
No diagnosis	6 (31.57%)
Medications prior to refer	
Antispasmodics	5 (26.31%)
Probiotics	5 (26.31%)
Valproate	3 (15.78%)
Levetiracetam	3 (15.78%)
Termination of events after initial visit	4-12 months

Once the diagnosis was established, the parents were reassured and no further drug therapy was provided. The children were slowly weaned off the antiseizure drugs.

The children were followed up for 6-15 months by direct clinic visits and telephonic interview. But, nine of 19 children did not attend any follow up visits after they were counseled about the nature of condition. None of the remaining children had a recurrence of events during

follow up. No other developmental, neurological and behavioral abnormalities were noted on follow up.

DISCUSSION

Gratification, a Latin word, literally means desire, to feel pleasure, or happiness. Though often called a disorder, it is normal behavioral variant seen in early childhood in some children.^{2,3,7} There were individual case reports and few studies describing series of patients in the literature.¹⁻⁵ A true incidence of gratification phenomena is not available. The gratification phenomenon is seen mostly in females. Two studies have reported it in 37% and 21% of male children, respectively.^{1,3} Only two male children were seen in our series of 19 (10.6%) patients.

The common age at presentation is between 4 and 12 months.^{1,3,7} Nine of 19(47.36%) children had onset before 12months. This was similar to Nechay et al study, where 48.38% of children had their events before 1year of age.¹ But two studies have that 93% and 100% of children had age at presentation before 13months.^{3,8}

GP has variable clinical presentations in early childhood and failure to recognize these behaviors. However, the typical clinical features in children as reported by Yang et al include Onset after the age of 3 months and before 3 years, stereotyped episodes of variable duration, vocalizations with quiet grunting, facial flushing with diaphoresis, pressure on the perineum with characteristic posturing of the lower extremities, no alteration of consciousness, cessation with distraction, normal examination, normal laboratory studies.²

In the present study, typical features were found in 12 children, and other have various atypical features. Koul et al reported three new patterns which were previously reported seldom.⁸ GP could pose a diagnostic challenge in early childhood if the clinician is not aware of its possibility because unlike in adolescents, it does not usually involve manual genital manipulation and its clinical presentations are variable, therefore it could be wrongly diagnosed as epilepsy, non epileptic paroxysmal movement disorder, infant colic or even gastroesophageal reflux disease.^{1,2,9}

In the present series, six children were diagnosed as epilepsy, five children as infantile colic and two children with urinary tract infection (UTI). This observation is similar to that of Nechay et al in their review of 31 cases of masturbatory behavior in children found that majority of the patients were initially misdiagnosed with seizure disorder; while Fleisher and Morrison in their case series, reported movement disorder as the commonest initially diagnosis.¹ Koul et al also reported in their study that 7(24.1%) were diagnosed with epilepsy and receiving antiseizure drugs.⁸ Some individual case reports available that GP was misdiagnosed as epilepsy and treated with antiseizure drugs.^{10,11,12}

A detailed history, examination and video recording by parents are enough to make diagnosis. One of the most important symptoms is that the child may be stopped during gratification if distracted and also shows anger and annoyance when interrupted.¹ No investigations are required, though many of these children have been underwent numerous investigations before referral.

The exact mechanism of GP is poorly understood, but has been associated with self-tension, boredom, excitement, and lack of stimulation.¹³ There are suggestions that dopaminergic pathways or limbic system in the brain may be involved in pleasure sensing at this age.¹⁴ UTI and parasitic infestation causing local irritation of genitalia in these children have also been postulated. However, two of our children had UTI.

Once the diagnosis of GP is made, reassurance of the family is the key in management. The parents should be educated that this is a behavioral variation that tends to become a habit. They need to know that it is harmless, not painful, and reassured that it will eventually subside with time.

Attempts should be made to engage the child in other play activities which could redirect their attention from masturbatory activities. Referral to behavioral psychology may be beneficial for some parents.¹⁰ In our series, all followed up children had remission without any therapy and none of the children referred for behavioral psychology.

Little data is available in the literature regarding long term follow-up of children with GP. Most of the children get remission by 2 to 3 years of age.³ One outcome study with two years follow-up revealed that infants rarely continue to have gratification phenomena beyond the age of two years.¹⁵ In this study, 10 children followed for 6 to 15 months, complete remission was noted by 4 to 12months after the initial visit. No other developmental, neurological and behavioral abnormalities were noted on follow up. Some long term studies reported that a possible link GP, which could be an early feature or a predictor of ADHD.^{3,16,17}

To the best of our knowledge, this is the first study and the largest cohort of children with GP from India. This study had several limitations. First, this was a single center study. Second, small sample size and short follow up unable to represent all pediatric population. However, larger and long term prospective studies are needed to further study this condition and outcome.

CONCLUSION

Gratification phenomenon is a benign and self-limited condition disappearing by 2 to 3 years of age spontaneously. A detailed history with video clipping helps in diagnosing the phenomenon. Clinicians should be aware of this condition and its variable patterns otherwise

it could be misdiagnosed and underwent unnecessary investigations and treatment. Only assurance and counseling of the parents is the main stay of management.

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