EDITORIAL

Gratification, Masturbation or Paroxysmal Hyperkinetic Motor Syndrome of Infancy?

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In this issue of the Canadian Journal of Neurological Sciences, Jan et al draw attention to an intriguing paroxysmal motor phenomenon in infants labelled "gratification disorder," "infantile masturbation," "self-stimulation in infancy" or variations thereof.[1-7] Descriptions of the disorder dating back to Bakwin and earlier,[8] are generally similar and stereotypical; the crucial role for home video recordings is highlighted by the example from Jan et al.[1] Onset in most cases is under one year of age, often as early as one to five months, with the overwhelming majority being female. Examples have been reported from several parts of the world. The events can (i) occur in the prone, supine and lateral positions, (ii) happen occasionally or several times a day, (iii) last from a minute to hours, and (iv) stop with distraction; some infants apparently get upset if interrupted. Episodes often occur before falling asleep, when the infants are described as anxious or bored, but never during sleep. A stressful event or genito-urinary disorder coincided with the onset in 85% of 61 children in Unal's controlled study,[9] an important point in history and management. During episodes, the lower limbs are stiff, often adducted, and there may be rubbing of the thighs; if the infant is supine, rhythmic pelvic movements may occur; movements of the tongue, neck (twisting), and hands have also been reported.[6,10] Direct genital contact is generally absent. Irregular breathing, diaphoresis, grunting and other vocalisations are often associated. Infants may exhibit similar behavior in car seats, high chairs or on laps of caregivers, and young children may do so against objects such as dolls or chairs etc. Analogies have been drawn between these behaviors and orgasmic activities in adults.[8] The episodes have often been mistaken for abdominal pain or seizures, and "distinction between paroxysmal movement disorders and masturbatory behavior can be difficult."[6] In most,[1-6,8] these behaviors apparently resolve by three to four years of age, but this may be an observational bias; by this age, children are not only less supervised, but most have also developed a sense of socially appropriate public behaviors.

Here, we discuss the presumptions underlying the terms "masturbation" and "gratification," and propose an alternate syndromic descriptor. We define infants as those < one year of age; our remarks apply in particular to those younger than six months, and when the events do not occur exclusively with an object, in a high chair or car seat.

Childhood masturbation has been defined as "self-stimulation of the genitalia often to achieve an orgasm."[11] The notion that fetuses and infants can have "erotic feelings" and masturbate,[8] likely dates back to the 1900s, and to possible misinterpretation and mistranslation of Freud's concepts of "infantile sexuality."[12-15] Freud cautioned that "the demonstrability of sexual manifestations in infants," was "a matter of interpretation."[15] His views on infant sexuality encompassed "pleasurable activities far removed from any concerns with reproduction,"[13] activities such as sucking, cuddling, hugging etc. Martinson suggested that infantile sexuality be viewed under a "general conceptual rubric, such as the pleasure-pain principle."[12] Although penile erections and vaginal secretions in response to stimulation have been interpreted as sexual arousal in infants,[8,11] these are generally considered reflexive.[16] "Masturbatory" activities in fetuses,[17-19] are also open to other interpretations. The social "sexual behaviors" of immature primates cannot be extrapolated to the "self-stimulatory" behavior of the human infant, as Bakwin did.[8] Therefore, infant sexuality must be considered from the infant's "perspective," not the adult's.[12,14,20]

A community-based study of 2-12 year olds, strongly linked sexual behavior in children to that in the family;[21] children in this age group may imitate adult behavior but it is unlikely that those < two years have sufficient cognition to do so. In a commentary on Bakwin's paper,[8] Elkind wrote that while "evidence for self-stimulatory behavior on the part of infants and children is abundant and clear-cut," interpretation of such behavior from the adult "sense" is questionable.[22] Sexual behaviors, including masturbation, involve "a complex of feelings, attitudes and fantasies;" a "complex" that "must wait upon the onset of the child's physical, cognitive, and emotional maturity."[22] Hence, the concepts of adult sexuality (and the desire-excitement-orgasm cycle) do not apply to infants,[12,14,20] and the pleasure (soothing) that infants and young children (certainly those < four years) experience from self-stimulation of the thighs and genitals cannot be considered to be either erotic or masturbatory. However, a pleasurable act may become a habit.[1,8] Authors of some clinical reports, Jan et al included, have acknowledged that the "gratification disorder" may have a different basis from the masturbatory behavior seen later in life.[1,3,23] Understandably, many parents also have reservations about accepting "masturbation" as an explanation for the episodes.[1,3,4,6] The apparently entrenched views on masturbation and orgasm in fetuses and infants,[8,11] need critical re-appraisal.

"Self-stimulatory behavior" and "gratification disorder" seem more appropriate and acceptable than "infantile masturbation," but these terms also reflect opinion, and do not evoke an image of the motor behaviors that bring infants to medical attention. Nechay et al suggested "benign idiopathic infantile dyskinesia" as an alternative diagnostic label;[4] however, "dyskinesia" does not effectively capture the spectrum of motor behaviors that have been described, and implies dysfunction in basal ganglia related systems. We propose: "Paroxysmal hyperkinetic motor syndrome of infancy," as a precise descriptive term. "Hyperkinetic" meets current definitional requirements.[24] "Syndrome" not only

acknowledges some clinical differences between cases but also reminds the clinician to ensure accuracy of diagnosis, self-stimulatory behavior being one possibility.

Continued usage of "infantile masturbation" and "gratification disorder" should also be discouraged to avoid cognitive biases such as confirmation, diagnosis momentum etc., Croskerry warns us against; these biases perpetuate erroneous hypotheses, and can hinder consideration of alternative diagnoses.[25]

We echo Jan et al's call for prospective studies.[1] Although described by many authors as a variant of normal behavior, (i) only a few case series of "the syndrome" have been described; it is unlikely that most parents would accept the behavior as normal and not seek medical attention, (ii) to our knowledge, these behaviors have not been explicitly reported in community-based studies, and (iii) true masturbatory behaviors generally start later in childhood. Clinical features that require explanation include, (i) the striking female preponderance, (ii) the observation that not all experience pleasure or gratification: thus at least one infant (Case 1 Reference 3) complained "it hurt,"[3] and some cry during it,[2] (iii) Nechay et al's descriptions of one infant becoming cyanotic, another exhibiting lip smacking, and a third appearing frightened,[4] and (iv) prolonged duration of some episodes, some lasting several hours,[6] rather long for "masturbatory" or "self-stimulatory" behavior. Better understanding can contribute to diagnostic specificity and management. Therefore, we suggest the syndrome be re-examined from developmental psychosexual, social, cultural, and biological perspectives. Would Freud, the pediatric neurologist, have wanted otherwise?

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