Pleasure and Displeasure: Thunderclap Headache in a 13-Year-Old Boy

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Abstract
Primary headache associated with sexual activity (PHASA) is a rare headache syndrome characterized by an acute, maximally intense headache during sexual activity and/or orgasm. While rare, it is a diagnosis that is widely accepted in adults; but, scarcely documented in children and adolescents. We aim to highlight the diagnostic process of this interesting headache syndrome in the pediatric population and add to the small list of reported cases in this group. Herein, we describe the case of a 13-year-old boy who presented with thunderclap headaches (TCH) associated with sexual activity. While more commonly diagnosed in adults, PHASA should be considered in sexually active children, though more ominous diagnoses should also be contemplated prior to establishing this diagnosis.

Keywords
headache, adolescents, pediatric, migraine, sexual activity

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Introduction
Primary headache associated with sexual activity (PHASA) is a rare headache syndrome described in The International Classification of Headache Disorders 3rd Edition (ICHD-3) as a headache during or following sexual activity and/or orgasm (Table 1).¹ While the headache may build gradually as a dull ache with incremental sexual excitement, it may also be abruptly explosive with orgasm and warrants exclusion of secondary headaches. Though widely described as a headache syndrome in adulthood, only 6 adolescent cases have been documented in the literature.²⁻⁵ The cases range from 12 to 19 years of age, though all but one are 16 years or older.³ Herein, we present a case of a 13-year-old boy with a PHASA to further highlight its incidence in younger adolescents who may not be as comfortable or willing to bring attention to this condition.

Case
A 13-year-old ambidextrous White male with Tourette syndrome, attention deficit hyperactivity disorder (ADHD), depression, and oppositional defiant disorder presented with recurrent severe headaches in the setting of masturbation. His most recent headache occurred one day prior to presentation following an episode of masturbation. He described a sudden, severe, and sharp headache, “like a gunshot” arising immediately upon reaching orgasm, with bilateral retro-orbital pain extending to the vertex of the head. There was associated nausea, photophobia, phonophobia, and osmophobia. However, no numbness, weakness, visual, or language impairment were associated with the headache. His home medications included methylphenidate, aripiprazole, ziprasidone, fexofenadine, trazodone, and famotidine; none of which had been added or adjusted immediately prior to the onset of these headaches. His mother and older brother had a history of migraines.

On further history, he had not yet engaged in sexual intercourse and denied any prior headaches, including exertional headache, until onset of masturbatory practices 2 to 3 months prior. It was unclear whether a headache occurred with his first episode of masturbation, though he noted headaches

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were not present with each episode of masturbation and were milder than the recent headaches; headache duration was unclear. Over the previous 7 days, he had masturbated daily and experienced severe headaches with each episode. The pain would gradually diminish in intensity before returning to its peak intensity upon the next masturbatory event. Details on the speed of masturbation were not elicited.

He first presented to an outside emergency department, where, given the sudden onset and explosive nature of his headache, a computed tomography (CT) angiogram of the head did not reveal a vascular pathology, including vasoconstriction, hemorrhage, dissection, or aneurysm. CT head was also normal. Magnetic resonance imaging, magnetic resonance angiogram, magnetic resonance venogram, or conventional angiography were not performed. He received fluids, ibuprofen, and sumatriptan with submaximal relief before being discharged home with follow-up in our clinic.

Upon arrival to our institution, he had a normal neurologic exam, including a normal fundoscopic exam. He was given ketorolac 10 mg leading to complete resolution of the headache. However, following masturbation the next morning the headache returned, and he re-presented for further evaluation at our emergency department. He received a combination of normal saline, diphenhydramine, prochlorperazine, and morphine. The patient was subsequently admitted for observation and ketorolac was given. The following morning his headache and the associated symptoms had fully resolved. Indomethacin 50 mg daily as needed 30 min prior to sexual activity was initiated upon discharge. Unfortunately, he was lost to follow-up.

**Discussion**

PHASA is a well-established primary headache disorder in adults. Though it can be orgasmic or preorgasmic, these 2 presentations are thought to be the same disease and since the release of ICHD-3 have been categorized as such. These episodes are characterized by increasing headache intensity with incremental sexual excitement and/or abrupt explosive intensity at orgasm. At their most intense severity, headaches can last 1 min to 24 h, with milder symptoms lasting up to 72 h. Its prevalence in the general adult population is around 1% and disproportionately affects males with a ratio as high as 3:1. Prevalence in sexually active adolescent populations may be like that seen in adults, though it is difficult to ascertain with only 6 previous cases. However, this case is only the second reported in a patient younger than 16 years of age. PHASA has been associated with other concurrent headache disorders, such as benign exertional headache and migraines, only 2 of the adolescent cases have been free of prior headache history like our patient. There is no documented relation between PHASA and Tourette syndrome or ADHD.

While PHASA is a primary headache, one must exclude a secondary headache, with the first onset of headache with sexual activity as mandated by ICHD-3 criteria. Only 1 of the 6 available cases did not have brain or vascular imaging. Subarachnoid hemorrhage, cerebral venous sinus thrombosis (CVST), reversible CVST (RCVS), and arterial dissection, may also occur in the setting of sexual activity, and are the main differential diagnoses of TCH, though other considerations include an intracranial tumor, migraine, primary exercise headache, and PHASA.

The absence of characteristic venous thromboembolic risk factors made CVST less likely, and the negative CT head and angiogram argued against hemorrhage, aneurysm, and tumor. RCVS was of special consideration in this case due to recurrent TCH and the use of methylphenidate and trazodone, which have been associated with RCVS, in pediatric and adult patients, respectively. While RCVS is often associated with vasoactive and serotonergic drugs, it is more commonly seen in adults, women more than men, and often in settings of vasoactive stress (postpartum, vasoactive drug use). In pediatric cases of RCVS, however, 81% of cases are reported in males. Imaging may initially be negative and recurrent imaging is often warranted, as demonstrated by Singhal’s 139 RCVS cases at 2 academic centers which showed 55% of patients had normal imaging on admission and 81% subsequently developed brain lesions. Unfortunately, the patient was lost to follow-up and repeat imaging was unobtainable. However, given the headache’s explosive onset, stereotyped association with sexual activity, and negative imaging studies, ICHD-3 criteria for PHASA were met in our patient.

The mainstay of treatment is premedication with indomethacin or a triptan 30 min prior to sexual activity, although post-activity use of NSAIDs and triptans is also described. While trigger avoidance may be used to reduce the frequency, daily prophylaxis with beta blockers, topiramate, or calcium channel blockers has been efficacious in uncontrolled case reports. Spontaneous remissions are frequent and prophylaxis may only be necessary for 2 to 6 months. As such, trigger avoidance should be reserved for refractory cases to not discourage or hinder sexual health, especially in young adolescents who are developing and exploring their sexuality. Because patients may be lost to follow-up (2 of the 7 pediatric cases), it is important to provide reassurance to patients and families about the absence of an ominous secondary pathology and the possibility of spontaneous remission. Patients should also be encouraged to seek medical help without any hesitation should the headaches recur or continue with masturbation as potential treatment options are available.
Although PHASA is more commonly seen in adults, this case adds to the literature a young adolescent with PHASA to highlight its presence in patients younger than 16 years of age. It is imperative that clinicians be aware to elicit potential triggers, especially in an age group that may be less willing to volunteer information about their sexual practices. Younger adolescents may feel stigmatized about acknowledging physiologic sexual practices, such as masturbation, and therefore are less likely to be forthcoming if those practices are associated with complications or pain. This is especially relevant in many countries and cultures around the world where sexual topics are not openly discussed. Moreover, in areas of the world where resources such as imaging may be limited, it is imperative to highlight the clinical features of this headache disorder. However, when resources are available, head and vascular imaging should be performed to exclude a secondary headache.

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Ethical Approval
Not applicable, because this article does not contain any studies with human or animal subjects.

References
1. Headache Classification Committee of the International Headache Society (IHS). The international classification of headache disorders, 3rd edition. Cephalalgia. 2018;38(1):1-211. doi:10.1177/0333102417738202.
2. Chakravarty A. Primary headaches associated with sexual activity—some observations in Indian patients. Cephalalgia. 2006;26(2):202-207. doi:10.1111/j.1468-2982.2005.01027.x.
3. Evers S, Peikert A, Frese A. Sexual headache in young adolescence: a case report. Headache. 2009;49(8):1234-1235. doi:10.1111/j.1526-4610.2009.01498.x.
4. Gelfand AA, Goadsby PJ. Primary sex headache in adolescents. Pediatrics. 2012;130(2):e439-e441. doi:10.1542/peds.2011-2624.
5. Patel J, Rothner D. Primary headache associated with sexual activity in adolescents: illustrative cases. J Pediatr Neurol. 2021. doi:10.1055/s-0040-1721827.
6. Frese A, Eikermann A, Frese K, Schwaag S, Husstedt IW, Evers S. Headache associated with sexual activity: demography, clinical features, and comorbidity. Neurology. 2003;61(6):796-800. doi:10.1212/01.wnl.0000083988.98336.a3.
7. Rasmussen BK, Olesen J. Symptomatic and nonsymptomatic headaches in a general population. Neurology. 1992;42(6):1225-1231. doi:10.1212/wnl.42.6.1225.
8. Biehl K, Evers S, Frese A. Comorbidity of migraine and headache associated with sexual activity. Cephalalgia. 2007;27(11):1271-1273. doi:10.1111/j.1468-2982.2007.01381.
9. Coffino SW, Fryer RH. Reversible cerebral vasoconstriction syndrome in pediatrics: a case series and review. J Child Neurol. 2017;32(7):614-623. doi:10.1177/0883073817696817.
10. Jensen J, Leonard J, Salottolo K, McCarthy K, Wagner J, Bar-Or D. The epidemiology of reversible cerebral vasoconstriction syndrome in patients at a Colorado comprehensive stroke center. J Vasc Interv Neurol. 2018;10(1):32-38.
11. Velez A, McKinnie J. Reversible cerebral vasoconstriction syndrome: a review of recent research. Curr Neurol Neurosci Rep. 2013;13(1):319. doi:10.1007/s11910-012-0319-y.
12. Singhal A. Diagnostic challenges in RCVS, PACNS, and other cerebral arteriopathies. Cephalalgia. 2011;31(10):1067-1070. doi:10.1177/0333102411410084.
13. Frese A, Rahmann A, Gregor N, Biehl K, Husstedt I, Evers S. Headache associated with sexual activity: prognosis and treatment options. Cephalalgia. 2007;27(11):1265-1270. doi:10.1111/j.1468-2982.2007.01449.x.
14. Evans RW. Topiramate for the prevention of primary headache associated with sexual activity: the third and fourth case reports. Headache. 2020;60(8):1800-1802. doi:10.1111/head.13900.
15. Evans RW, Pascual J. Expert opinion: orgasmic headaches: clinical features, diagnosis, and management. Headache. 2000;40(6):491-494. doi:10.1046/j.1526-4610.2000.00075.x.