Transient sixth cranial nerve palsy following orgasm abrogated by treatment with sympathomimetic amines

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Summary

Purpose: To describe a unique disorder where a transient 6th nerve palsy leading to diplopia following orgasm developed in a 28-year- old woman. This coincided with a weight gain of 100 pounds in a short time without a corresponding change in dietary habits. *Materials and Methods:* She was treated with the sympathomimetic amine dextroamphetamine sulfate. *Results:* Indeed she immediately responded to treatment with dextroamphetamine sulfate sustained release capsules with complete resolution of the episodes of 6th nerve palsy following orgasm. *Conclusions:* The main importance of this case is that it suggests that orgasm causes a transient generalized decrease in sympathetic nervous system activity and that the achievement of an orgasm may require an increase in the sympathetic nervous system activity.

Key words: Sixth cranial nerve palsy; Diplopia; Orgasm; Sympathetic nervous system hypofunction; Dextroamphetamine sulfate.

Introduction

Clinical issues regarding orgasm can be multifold, e.g., failure to achieve one, the need for excessive orgasm, and severe pelvic pain following orgasm.

The authors report here a unique complication of orgasm in a woman – a sixth nerve palsy associated with diplopia. Even more importantly they report a unique pharmacological therapy that completely corrected the problem.

The fact that the therapy used a sympathomimetic amine which has been used to treat a variety of disorders characterized by diminished sympathetic nervous system activity implies that orgasm may result in a transient hypofunction of the sympathetic nervous system.

Case Report

A 28-year-old woman complained of episodes where her left eye would suddenly turn inward and she would see double. These episodes lasted only a few minutes in duration and were associated with times when she would stand up quickly. Within a few weeks of onset, the diplopia began occurring with every orgasm even though spontaneous occurrences weaned. Restoration of normal vision would return in about 15 minutes.

At first the patient thought that the problem could be a form of vertigo related to an inner ear problem. However, a specialist in otolaryngology did not know the cause and referred her to a neurologist. As it progressed, the episodes occurred more frequently and appeared to be associated with certain activities such as positional changes when she would go from sitting to standing or bending over. She also noticed they were more frequent premenstrually. However, she said without fail it would always happen with an orgasm and that these lasted longer than the spontaneous episodes.

She was referred to a neuro-opthalmologist who performed a lumbar puncture which was also negative. The neurologist ordered an electroencephalogram (EEG), magnetic resonance imagining (MRI) of the brain, and a magnetic resonance artery (MRA) study but all were normal. The diagnosis was a sixth nerve palsy diagnosed by clinical observation by a neuro-opthalmologist following a self-induced orgasm. None of the specialists had any suggestions for etiology or for management. The only suggestion was not to have an orgasm for fear of a possible cerebrovascular accident. Their other suggestion was to seek the opinion of an endocrinologist, particularly a reproductive endocrinologist, which is how the present authors first had their chance to evaluate this woman with this fascinating medical history.

Taking a further history, the patient had a 100 pound weight gain without any subsequent increase in her diet. Furthermore she had developed such severe backache with menstruation that her husband had to help her out of bed during that time.

On physical examination she was six feet tall, her weight was 285.5 pounds, blood pressure 120/70, thyroid normal size and non-tender, heart rate 80 and regular, no murmurs or gallops, no hepatosplenomegaly or abdominal tenderness, and no obvious pitting edema of the extremities.

Laboratory studies showed thyroid studies were normal including free thyroxin level and TSH (the latter at 1.26 mIU/ml). A water load test was abnormal with 1,750 ml urine excretion after four hours while supine but only 500 ml after standing for four hours (abnormal < 55% excretion) [1]. A 1,500 ml water load over 30 minutes was ingested initially each day prior to measuring urine excretion.

Based on the results of the abnormal water load test, the authors concluded that the inappropriate weight gain was most likely related to a defect in the sympathetic nervous system leading to marked fluid retention especially in the upright posture [1-3]. The defect was found to be related to an inappropriately weak response from the sympathetic nervous system to diminish capillary permeability and to prevent the transudation of fluid from intravascular to extravascular spaces that would occur from the increase in hydrostatic pressure that occurs in the orthostatic position [4].

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She was advised that she may lose weight following treatment with the sympathomimetic amine, dextroamphetamine sulfate which seems to diminish capillary permeability when standing and thus inhibits edema [5]. She was also advised that though there was no precedent for her specific orgasm condition, based on the wide variety of disorders that were corrected or alleviated by treatment with dextroamphetamine sulfate, it would not be surprising if the strange sixth nerve palsy with and without orgasm may similarly improve [6].

After one month of taking dextroamphetamine sulfate ten mg extended release capsules upon awakening and ten mg at noon the woman reported significant improvement in her lower back pain. The use of sympathomimetic amines has also been demonstrated to provide immediate relief from backache in some cases that were refractory to other therapies [7]. The spontaneous episodes of sixth nerve palsy which previously happened several times per day were reduced to just twice in the month and were also reduced in duration. Interestingly she reported complete resolution of the sixth nerve palsy episodes occurring with orgasm which previously occurred 100% of the time after each orgasm. She lost 9.5 pounds in that first month.

After one year of therapy she reported no episodes of diplopia following orgasm. The one exception was the two times she ran out of her prescription for a few days. She has lost 55 pounds so far.

Discussion

This disorder of diminished sympathetic nervous activity seems to be responsible for a large variety of chronic debilitating disorders not amenable to other therapies including edema, urticaria, chronic lower abdominal pain, interstitial cystitis, pelvic pain, backaches, esophageal motility disorders, gastroparesis, arthritis, and vasomotor symptoms [6, 8]. This has led to the hypothesis that the condition underlying these symptoms is either related to the effects of edema or more likely, to the absorption of toxins into epithelial cells related to increased cellular permeability secondary to diminished sympathetic nervous system activity as evidenced by quick and sustained response to the sympathomimetic amine dextroamphetamine sulfate [6, 8].

Several case reports suggest that the main etiology of the pain and other symptoms may be more associated with the absorption of toxins and chemicals into tissues because of a diminished sympathetic nervous system activity that is not properly suppressing cellular permeability. For example, a cure of pseudointestinal obstruction with dextroamphetamine sulfate therapy was not associated with weight gain and edema but in fact with marked weight loss down to a life threatening level. Soon after starting dextroamphetamine sulfate, her abdominal pain and early satiety markedly improved, as did her bowel movements and she quickly lost weight to the proper level [9].

Other areas of gastrointestinal motility such as the esophagus or stomach seem to be prone to defects in diminished sympathetic activity and respond to therapy with sympathomimetic amines when all other therapies were ineffective [10, 11]. There is also evidence that therapy with sympathomimetic amines can markedly and quickly improve the symptoms of severe long-standing Crohn's disease and ulcerative colitis that were refractory to standard therapies [12, 13].

Evidence that defects in the sympathetic nervous system can lead to skeletal muscle abnormalities is evidenced by marked improvement in chronic fatigue syndrome with treatment with sympathomimetic amines [14]. Improvement in other cerebral conditions was illustrated by the marked improvement of headaches, including migraines with sympathomimetic amine therapy, which had been refractory to other medications [15, 16].

Based on these observations, one hypothesis to explain this very unusual syndrome reported here of sixth nerve palsy with orgasm is that the palsy was related to the absorption of toxins into the sixth cranial nerve or the oculomotor muscles that it innervates. The authors hypothesize that there was usually sufficient sympathetic tone to prevent diplopia except under certain circumstances, e.g., sitting up too quickly. However they hypothesize the process of orgasm may be normally followed by a period of decreased sympathetic nervous system activity which would further allow an increase in cellular permeability leading to a critical concentration of toxins and chemicals to cause temporary muscle paresis. Some additional local defect of these muscles must have also been present causing their susceptibility to a temporary decrease in sympathetic tone.

A search of the literature failed to reveal a similar report. The case then also presents another unique presentation for the relatively common problem in women with sympathetic nervous system hypofunction referred to as the sympathetic neural hyperalgesia edema syndrome [8]. Specifically this case presents the report of a strange association of diplopia related to temporary sixth nerve paresis following orgasm amenable to therapy with sympathomimetic amines.

The universal occurrence of the sixth nerve palsy following orgasm and the complete correction of this problem following treatment with a sympathomimetic amines strongly suggests that orgasm is followed by a transient decrease in sympathetic nervous system activity. However, in most people without this defect of sympathetic nervous system hypoactivity, the transient decrease in sympathetic tone does not cause symptoms because there is no presence of a target tissue that has another defect and is thus prone to malfunction following diminished sympathetic activity. Since most cases of the sympathetic neural hyperalgesia edema syndrome are not associated with diplopia following orgasm, this woman presumably also had some rare defect in either the sixth nerve or the extraocular muscles per se allowing the temporary paresis and subsequent diplopia to occur.

References

[1] Thorn G.W: "Approach to the patient with idiopathic edema or periodic swelling". *JAMA*, 1968, *206*, 333.

[2] Kuchel O., Horky K., Gregovova I., Marck J., Kopecka J., Kobilkova J.: "Inappropriate response to upright postures: a precipitating factor in the pathogenesis of idiopathic edema". *Ann. Intern. Med.*, 1970, *73*, 24552.

[3] Edward O.M., Bayliss R.I.S.: "Idiopathic edema of women: clinical and investigative study". *Q.J. Med.*, 1976, *177*, 125.

[4] Streeten D.H.P.: "Idiopathic edema: pathogenesis, clinical features and treatment". *Metabolism*, 1978, *27*, 353.

[5] Check J.H., Shanis B.S., Shapse D., Adelson H.G.: "A randomized comparison of the effect of two diuretics, a converting enzyme inhibitor and a sympathomimetic amine on weight loss in die-refractory patients". *Endoc. Prac.,* 1995, *1*, 323.

[6] Check J.H., Katsoff D., Kaplan H., Liss J., Boimel P.: "A disorder of sympathomimetic amine leading to increased vascular permeability may be the etiologic factor in various treatment refractory health problems in women". *Med. Hypoth.*, 2008, *70*, 671.

[7] Check J.H., Wilson C., Cohen R.: "A sympathetic nervous system disorder of women that leads to pelvic pain and symptoms of interstitial cystitis may be the cause of severe backache and be very responsive to medical therapy rather than surgery despite the presence of herniated discs". *Clin. Exp. Obst. Gyn.*, 2011, *38*, 175.

[8] Check J.H., Cohen R., Katsoff B., Check D.: "Hypofunction of the sympathetic nervous system is an etiologic factor for a wide variety of chronic treatment-refractory pathologic disorders which all respond to therapy with sympathomimetic amines". *Med. Hypoth.*, 2011, *77*, 717.

[9] Check J.H., Cohen R.: "Successful treatment of a female with chronic pseudo-intestinal obstruction with sympathomimetic amines and thyroid hormone replacement". *Clin. Exp. Obst. Gyn.*, 2010, *37*, 115.

[10] Leskowitz S.C., Shanis B.S., Check J.H.: "Resolution of atypical chest pain during treatment for idiopathic orthostatic edema". *Am. J. Gastroenterol.*, 1990, *85*, 621.

[11] Boimel P., Check J.H., Katsoff D.: "Sympathomimetic amine therapy may improve refractory gastroparesis similar to its effect on chronic pelvic pain – case report". *Clin. Exp. Obstet. Gynecol.*, 2007, *34*, 185.

[12] Check J.H., Katsoff B., Cohen R.: "A novel highly effective medical treatment of severe treatment refractory Crohn's disease using sympathomimetic amines – case report". *Inflam. Bowel Dis.*, 2010, *16*, 1999.

[13] Check J.H., Katsoff B., Cohen R.: "A case report showing that a woman with ulcerative colitis refractory to standard therapy responded well to the sympathomimetic amine dextroamphetamine sulfate". *Inflam. Bowel Dis.*, 2011, *17*, 870.

[14] Check J.H., Cohen R.: "The sympathetic neural hyperalgesia edema syndrome, a frequent cause of pelvic pain in women, mistaken for lyme disease with chronic fatigue". *Clin. Exp. Obst. Gyn.*, 2011, *38*, 412.

[15] Check J.H., Check D., Cohen R.: "Sympathomimetic amine therapy may markedly improve treatment resistant headaches related to a vascular permeability defect common in women – presentation of two cases". *Clin. Exp. Obst. Gyn.*, 2009, *36*, 189.

[16] Check J.H., Cohen R.: "Marked improvement of headaches and vasomotor symptoms with sympathomimetic amines in a woman with the sympathetic hyperalgesia-edema syndrome". *Clin. Exp. Obst. Gyn.*, 2011, *38*, 88.

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