

CASE REPORT

Persistent sexual arousal in a woman with associated cardiac defects and raised atrial natriuretic peptide

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Summary: The persistent sexual arousal syndrome (PSAS) is a newly described entity where the woman becomes involuntarily genitally aroused for extended periods of time in the absence of sexual desire and is distressed by this situation. The cause of this sexual problem is not well understood. We describe such a case where the subjective feelings were confirmed by observing genital engorgement. In her case, PSAS came on after initiation of fludrocortisone given for hypotension and bradycardia that was associated with an atrial septal defect (ASD). We argue that the combined effect of the ASD and fludrocortisone may be associated with an increase in her levels of atrial natriuretic peptide (ANP). ANP causes profound vasodilation and vascular leakage. We postulate that the high serum levels of ANP in her case may be contributory to her PSAS.

Keywords: persistent sexual arousal syndrome (PSAS), female sexual dysfunction, atrial septal defect (ASD), atrial natriuretic peptide (ANP)

INTRODUCTION

Persistent sexual arousal syndrome (PSAS) describes a new entity where a woman becomes involuntarily genitally aroused for extended periods of time in the absence of sexual desire.¹

Leiblum and Nathan² first described persistent genital arousal in women in 2002. In 2003, PSAS in women was included into the nomenclature of female sexual dysfunction.³ A more recent internet survey of women with PSAS found that the commonest features of the syndrome were: involuntary genital and clitoral arousal persisting for extended periods, genital arousal unrelated to subjective feelings of sexual desire, and genital arousal not relieved with orgasms.⁴ The aetiology of the syndrome is unknown, although individual case reports so far suggest that local arteriovenous fistulas and high intake of soy products can cause the syndrome in individual patients.⁵

We report a case of PSAS in a woman with a patent foramen ovale (PFO) and an increase in atrial natriuretic peptide (ANP).

CASE

We describe a 57-year-old woman; her past medical history includes benign breast cysts and a hysterectomy plus

unilateral oophorectomy aged 32 years for menorrhagia. In 2001, she was admitted with symptoms of severe hypotension (BP 75/30) and was found to have two atrial septal defects (ASDs). Bradycardia of 40 bpm necessitated cardiac pacemaker insertion. In November 2004, she started fludrocortisone, because of symptomatic hypotension, which subsequently was stopped because it gave no symptomatic relief. Since December 2004, she experienced spontaneous and prolonged feelings of genital arousal alleviated briefly by sexual intercourse, masturbation or use of a vibrator. Her sexual problem causes great distress, and she describes being constantly on the verge of orgasm. Examination of her genitalia showed oedematous, vasoengorged labiae, vulva and clitoris.

She has been extensively investigated by a number of specialist clinicians. Investigations include electro-encephalogram, computer tomography (CT) brain, CT spinal cord, myocardial perfusion scan, pulmonary function tests, pelvic ultrasound, lumbar puncture and nerve conduction studies, which were all normal. Autonomic function tests indicated a low supine blood pressure with no evidence of orthostatic hypotension and no evidence of cardiovascular autonomic failure. A 24-hour ambulatory blood pressure and heart rate profile showed a low daytime supine blood pressure with an expected circadian fall. Individual scrutiny of the data showed that her heart rate rose to a greater extent than expected at times when exerting herself minimally, although not in the region expected of the postural tachycardia syndrome. A modified exercise tolerance test evoked a marked heart rate response, and

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clonidine suppression testing was normal. Plasma adrenaline and noradrenaline were normal.

CT coronary angiogram demonstrated her left superior vena cava draining into her coronary sinus and right superior vena cava draining into the right atrium (a recognized anatomical variant). A trans-oesophageal echo showed a large coronary sinus and large PFO, a fossa ovalis was seen on the intra-atrial septum and a flap identified. The right heart was morphologically normal on echo, and the Atrial and Ventricular pacing device was shown to be functioning appropriately.

Investigations have also excluded a neoplastic or paraneoplastic-type syndrome. Sex hormone profile was consistent with a pre-menopausal state. Androgen, pituitary and adrenocortical hormone screening was normal. Her serum level of ANP was 18.6 pg/mL (mean of 6 assays on one serum sample) (upper limit normal 8.8 pg/mL).

Symptomatically, she has gained no relief using diazepam, gabapentin, carbamazepine, paroxetine and pregabalin. Some initial improvement occurred with 50 mg of amitriptyline, but the genital symptoms soon relapsed. Currently, she takes midodrine; she feels generally better, her BP remains low (90/50), but genital arousal persists unalleviated. Unfortunately, the cardiologist's opinion was that the risk of percutaneous closure of the PFO outweighed the potential benefits. We are currently thinking of alternative managements, and providing her with symptomatic support including pharmacotherapy and cognitive behaviour therapy.

DISCUSSION AND CONCLUSION

It has been suggested that a cause of migraine is shunting of blood from the right to left atrium in patients with atrial septal defects (ASDs).⁶ In such patients, there is pathological right atrial hypertrophy. The stretched right atrium responds by secreting increased amounts of ANP. Fludrocortisone has also been shown to increase ANP.⁷

ANP causes vasodilatation via raised intracellular guanylate cyclase and cyclic GMP stimulation, increasing

vascular endothelial permeability, all of which could result in local genital vasocongestion and oedema, resulting in a state of PSA. Closure of ASDs has been shown to cause significant decreases in ANP and relief of migraine.⁸

We report a case of a woman with persistent sexual arousal and a PFO. It may well be that the PSA and cardiological abnormalities exist coincidentally in her case. However, it is possible that the PFO combined with fludrocortisone resulted in abnormally elevated circulating ANP levels, thus leading to a state of PSA and that this, at least in part, contributed to her genital engorgement and symptomatic persistent genital arousal. Why raised levels of ANP in the systemic circulation should only cause genital problems appears as enigmatic to us as why it should only cause migraine in other patients.

REFERENCES

- 1 Goldmeier D, Leiblum SR. Persistent genital arousal in women – a new syndrome entity. *Int J STD AIDS* 2006;**17**:215-16
- 2 Leiblum S, Nathan S. Persistent sexual arousal syndrome in women: a not uncommon but little recognised complaint. *J Sex Relationship Ther* 2002;**17**:191-8
- 3 Basson R, Leiblum S, Brotto L, et al. Definitions of women's sexual dysfunctions reconsidered: advocating expansion and revision. *J Psychosomatic Obstet Gynecol* 2003;**24**:221-9
- 4 Leiblum S, Brown C, Wan J, Rawlinson L. Persistent sexual arousal syndrome: a descriptive study. *J Sexual Med* 2005;**2**:331-7
- 5 Amsterdam A, Abu-Rustum N, Carter J, Krychman M. Persistent sexual arousal syndrome associated with increased soy intake. *J Sexual Med* 2005;**2**:338-40
- 6 Wilmshurst PT, Pearson MJ, Nightingale S, Walsh KP, Morrison WL. Inheritance of persistent foramen ovale and atrial septal defects and the relation to familial migraine with aura. *Heart* 2004;**90**:1315-20
- 7 Cohen N, Gilbert R, Wirth A, Casley D, Jerums G. Atrial natriuretic peptide and plasma renin levels in assessment of mineralocorticoid replacement in Addison's disease. *J Clin Endocrinol Metab* 1996;**81**: 1411-15
- 8 Muta H, Ishii M, Maeno Y, et al. Quantitative evaluation of the changes in plasma concentrations of cardiac natriuretic peptide before and after transcatheter closure of atrial septal defect. *Acta Paediatrica* 2002;**91**: 649-52

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