

Comorbidity of Anorexia Nervosa in a Male Patient with Congenital Hypogonadism: Case Report

Mustafa Ozten¹, Atila Erol²

¹Psychiatry Resident, ²Prof. Dr., Sakarya University Research and Training Hospital, Department of Psychiatry, Sakarya - Turkey

ABSTRACT

Comorbidity of anorexia nervosa in a male patient with congenital hypogonadism: case report

Male hypogonadism is a result of insufficient testicular androgen production. In addition to low circulating testosterone levels, patients develop clinical symptoms such as osteopenia, decrease in muscle mass, libido, energy and stamina, decline in cognitive function, increased adiposity, and erectile dysfunction. Anorexia nervosa (AN) is quite rarely seen in men, being usually ten times less common than in women, and shows differences in many aspects, from causation to presentation. As AN is connected to hunger physiology, it entails significant medical complications affecting, as in any general medical condition, a number of systems, including the genitourinary system. It is known that a prolonged manifestation of AN or cachexia may cause hypogonadism. This article presents the case of a patient with untreated hypogonadism, diagnosed in his infancy, who at the age of 18 developed AN.

Key words: Anorexia nervosa, hypogonadism, male



ÖZET

Konjenital hipogonadizmi olan erkek hastada ek hastalık olarak anoreksiya nervoza: Olgu sunumu

Erkek hipogonadizmi testisin androjen üretme yetersizliğine bağlı oluşmaktadır. Hastalarda, dolaşımda düşük testosteron seviyeleri yanında osteopeni, kas kütlesi, libido, enerji ve dayanıklılıkta azalma, bilişsel işlevlerde düşüş, yağ miktarında artış ve erektil işlev bozukluğu gibi klinik belirtiler gözlenir. Anoreksiya nervoza (AN) erkeklerde oldukça nadir görülen, genelde kadınlardan on kat daha az görülen ve ortaya çıkış nedenlerinden seyrine kadar pek çok alanda farklılıklar gösteren bir durumdur. Açlık fizyolojisine bağlı olarak AN da önemli tıbbi komplikasyonlar gelişmektedir ve bu genel bir tıbbi hastalık gibi pek çok sistemi etkilemektedir. AN'de etkilenen sistemlerden biri de genitouriner sistemdir; uzun süren AN tablosunun veya kaşeksinin hipogonadizme neden olabileceği bilinmektedir. Bu yazıda bebeklikten itibaren hipogonadizm teşhisi olan ve tedavi almayan, 18 yaşında iken AN gelişen bir vaka sunulmaktadır.

Anahtar kelimeler: Anoreksiya nervoza, hipogonadizm, erkek

Address reprint requests to / Yazışma adresi: Mustafa Ozten, Sakarya University Research and Training Hospital, Department of Psychiatry, Adnan Menderes Cad. Sağlık Sok. No: 195 54100 Adapazarı/Sakarya, Turkey

Phone / Telefon: +90-264-255-2106

E-mail address / Elektronik posta adresi: drozten@gmail.com

Date of receipt / Geliş tarihi: September 26, 2013 / 26 Eylül 2013

Date of acceptance / Kabul tarihi: March 5, 2014 / 5 Mart 2014

INTRODUCTION

Male hypogonadism is defined as testicular insufficiency in testosterone secretion, production of spermatozoa, or both. In addition to low testosterone levels, patients develop clinical symptoms such as osteopenia, increased adipose tissue, decrease in muscle mass, energy and stamina, loss of cognitive function, decreased libido and erectile dysfunction.

Anorexia Nervosa (AN) is an eating disorder more common in young girls, rarely seen in men, and if left untreated, carries a high mortality risk. It occurs 5-10 times more frequently in women than in men (1-2);

one study found that only 4% of all patients suffering from AN or bulimia were males (3). It is accepted that social, genetic, and psychological factors, personality traits and trauma history play a role in the development of eating disorders. Furthermore, steroid hormones affect eating behavior and the development of eating disorders.

Important medical complications are developing in AN; in particular, the levels of prolactin, thyroid hormones, gonadal hormones and, in men, testosterone are changing (4,5). Most of the endocrine changes in AN are a physiological adaptation against hunger, and they are generally reversible with weight gain.

In this article, a case is presented of a man with untreated hypogonadism, diagnosed in his infancy, who at the age of 18 developed AN.

CASE

O.N. 22-year-old male patient, primary school graduate, single, unemployed, living with his family. Complaints include weight loss, lack of appetite, eating little at meals, often skipping meals altogether, getting angry when asked to eat, fear of gaining weight and imagining to be overweight. Height 175cm, weight 52kg (body mass index [BMI]=16.9). His medical history included congenital hypogonadism, right testicular agenesis, hypoplasia of the left testis.

The patient was first admitted to hospital at the age of 8 for diagnosis and treatment, where right testicular agenesis, left testis hypoplasia and congenital hypogonadism were found. While luteinizing hormone (LH), follicle stimulating hormone (FSH) and total and free testosterone were low, other hormone and blood values were normal. Brain imaging showed no pathologies in the hypothalamic and pituitary regions, and the patient was discharged with recommendation for testosterone treatment and follow-up consultations. However, the patient did not use the testosterone treatment and did not attend the follow-up. At age 16, minimal hair growth began in the arm pits and the pubic region, but he had neither ejaculations nor erections, secondary sexual characteristics did not develop, and a chronic reduction of libido was stated. At the age of 18, reduced eating and sleeping, fear of gaining weight, feeling overweight, skipping meals and expressions of anger and irritation when asked to eat began. In that period, the patient did not want to eat, and when asked for the reasons, he said he did not feel hungry and did not need to eat, and if he did, he would gain too much weight and feel sick to his stomach; this nausea would disturb him and he did not want to experience this. Recently, he stopped having breakfast, just eating a few spoons of home-cooked food at lunch and dinner, which made him feel full. Although the patient did not vomit nor used weight loss drugs and did not exercise excessively, he began to lose weight rapidly, going from

60kg to 46kg in 6 months (BMI=15.02) and was admitted to our clinic. Other than his hypogonadism, the patient's medical history revealed no abnormalities; nor did his family history. He had a good relationship with his family and friends and showed no difficulties in communicating with other people. He was quite weak for his age, extroverted and had good communication skills. He said that he did not want to talk about food and hypogonadism, did not think about these issues and had plans for his future. With this history and symptoms, the patient was diagnosed with restrictive type AN. LH, FSH, and testosterone levels were measured again and found to be low. The patient was started on cyproheptadine hydrochloride suspension 3x1 and mirtazapine 30mg/day. The patient took the drugs regularly and within 2 months increased his weight to 53kg. At the end of the second month, when the patient's symptoms had been partly resolved, he stopped drug use and follow-up. One year later, the patient again presented to our outpatient clinic, saying his eating and sleeping had returned to normal and his weight was stable; he did not want to gain more weight or take any medication. While the hypogonadism-related symptoms were continuing, he would not accept any treatment. His weight was measured as 52kg (BMI=16.9), treatment was recommended, but he did not accept.

DISCUSSION

Male hypogonadism is due to insufficient production of testicular androgen. The cause can be primary or secondary, but either way, the clinical signs are usually identical. In their metabolism, men with low androgen levels have high concentrations of glucose and insulin and an elevated rate of obesity with increased incidence of type 2 diabetes (6,7). In our hypogonadic case, the development of AN does not accord with the assumption that hypogonadism comes with a higher risk of weight gain and obesity (8,9).

Difficult and traumatic life events are known to play an important role in the development of AN, and it has been emphasized that in males psychosocial life events have a more prominent place in the

emergence of AN than in females (10). The medical problems and treatment difficulties experienced by our patient are thought to have played a role in the emergence of AN.

Families of anorexic patients have been described as close-knit, overprotective, intrusive, with problems especially in the field of conflict resolution (11,12). Our patient's family could be characterized as well balanced, exhibiting a positive attitude and sensitivity to the child's needs, communicating warmly, being flexible, tolerant and supportive. However, during his medical problems, the patient showed anger towards his family because of the failure of treatment. Our patient's family structure is not considered to have contributed to the development of his AN.

Adolescence is a phase of life associated with growth from the perspective of emphasizing sexuality in relationships and taking an initial step towards identity formation. One of the accepted opinions is that cases of AN during adolescence have difficulties in their identity formation stage, rejecting it, preferring to have a child's body (1,2,13-16). It has been reported that in male AN cases homosexual and asexual behavior and disorders related to sexual roles are more common than on average in society (17,18). In our case, it could be thought that symptoms caused by medical problems, such as the failure to develop secondary sexual characteristics and the absence of ejaculation and erection caused difficulties in the sexual identity formation stage, leading to the patient refusing to grow up and not wanting to eat.

Steroid hormones influence eating behavior and the development of eating disorders. Studies indicate that abnormalities in anorexigenic and orexigenic hormones affect eating patterns, energy balance and bone mass (19). It is known that ghrelin and neuropeptide Y (NPY) increase the appetite, while leptin reduces it. Ghrelin has been reported to be inversely correlated with the androstenedione and testosterone levels (20,21). It has been shown that androgens inhibit the production of leptin (22,23). Low leptin levels can lead to hypothyroidism, hypercortisolism, osteopenia, changes in the immune system, amenorrhea in women, and increased activity. In our case, it can be assumed that low androgen levels, by way of affecting hormones influencing the appetite, had an impact on eating behavior and played a role in the etiology of the developing eating disorder.

Russ et al. (24) and Wesselius et al. (25) reported that the neuroendocrine effects of AN in male patients caused hypogonadism and that the disorder could be normalized through weight gain. However, to this day only few cases with congenital hypogonadism and anorexia nervosa have been reported. In a case of congenital hypogonadism diagnosed with AN, Ohzeki et al. (26) have tried to explain the role of sex hormones in AN. In our literature review, not a single case report with these characteristics was found in our country. Our case importantly demonstrates that hypogonadism, which may appear as a complication occurring in AN, may in itself constitute a risk factor for the development of AN.

REFERENCES

1. Kaya N, Cilli AS. Anorexia nervosa. *General Medical Journal* 1997; 7:107-110. (Turkish)
2. Aydemir O, Koroglu E. Eating Disorders: In Koroglu E (Translation Editor). *DSM-IV Diagnostic and Statistical Manual of Mental Disorders*, 4th. Edition. Ankara: Hekimler Yayin Birliđi, 1998, 727-746. (Turkish)
3. O'Brien J, Patrick A. Cost of the acute hospitalization for anorexia nervosa and bulimia. *Value in Health* 2001; 4:146-152.
4. Tomova A, Kumanov P. Sex differences and similarities of hormonal alterations in patients with anorexia nervosa. *Andrologia* 1999; 31:143-147.
5. Klapper F, Gurney VW, Wiseman CV, Cheng HT, Halmi KA. Psychiatric management of eating disorders. *Nutr Clin Care* 1999; 2:354-360.
6. Grossmann M, Thomas MC, Panagiotopoulos S, Sharpe K, Macisaac RJ, Clarke S, Zajac JD, Jerums G. Low testosterone levels are common and associated with insulin resistance in men with diabetes. *J Clin Endocrinol Metab* 2008; 93:1834-1840.
7. Ding EL, Song Y, Malik VS, Liu S. Sex differences of endogenous sex hormones and risk of type 2 diabetes: a systematic review and meta-analysis. *JAMA* 2006; 295:1288-1299.

8. Clement K, Vaisse C, Lahlou N, Cabrol S, Pelloux V, Cassuto D, Gourmelen M, Dina C, Chambaz J, Lacorte JM, Basdevant A, Bougneres P, Lebouc Y, Froguel P, Guy-Grand B. A mutation in the human leptin receptor gene causes obesity and pituitary dysfunction. *Nature* 1998; 392:398-401.
9. Goldstone AP, Holland AJ, Hauffa BP, Hokken-Koelega AC, Tauber M; speakers contributors at the Second Expert Meeting of the Comprehensive Care of Patients with PWS. Recommendations for the diagnosis and management of Prader-Willi syndrome. *J Clin Endocrinol Metab* 2008; 93:4183-4197.
10. Rosen DS. Eating disorders in children and young adolescents: etiology, classification, clinical features and treatment. *Adolesc Med* 2003; 14:49-59.
11. Lai KY, Pang AH, Wong CK. Case study: early-onset anorexia nervosa in a Chinese boy. *J Am Acad Child Adolesc Psychiatry* 1995; 34:383-386.
12. Isik Y, Gokler B. Psychodynamics of anorexia nervosa. *Psychiatry in Turkiye* 2001; 3:10-15. (Turkish)
13. Tomova A, Kumanov P. Sex differences and similarities of hormonal alterations in patients with anorexia nervosa. *Andrologia* 1999; 31:143-147.
14. Klapper F, Gurney VW, Wiseman CV, Cheng HT, Halmi KA. Psychiatric management of eating disorders. *Nutr Clin Care* 1999; 2:354-362.
15. Maner F. Eating Disorders. *Psikiyatri Dünyası* 2001; 5:130-139. (Turkish)
16. Canat S. Eating Disorders: In Gulec C, Koroglu E (editors). *Basics of Psychiatry*. Ankara: Hekimler Yayin Birliđi, 1998; 661-672. (Turkish)
17. Johnson ME, Brems C, Fischer P. Sex role conflict, social desirability, and eating-disorder attitudes and behaviors. *J Gen Psychol* 1996; 123:75-87.
18. Oleinikov AN. Specific factors of sex behavior in patients with anorexia nervosa and bulimia. *Zh Nevrol Psikhiatr Im S S Korsakova* 2000; 100:19-22. (Russian)
19. Inui A. Eating behavior in anorexia nervosa -- an excess of both orexigenic and anorexigenic signaling? *Mol Psychiatry* 2001; 6:620-624.
20. Pagotto U, Gambineri A, Vicennati V, Heiman ML, Tschöp M, Pasquali R. Plasma ghrelin, obesity, and the polycystic ovary syndrome: correlation with insulin resistance and androgen levels. *J Clin Endocrinol Metab* 2002; 87:5625-5629.
21. Glintborg D, Andersen M, Hagen C, Frystyk J, Hulstrom V, Flyvbjerg A, Hermann AP. Evaluation of metabolic risk markers in polycystic ovary syndrome (PCOS). Adiponectin, ghrelin, leptin and body composition in hirsute PCOS patients and controls. *Eur J Endocrinol* 2006; 155:337-345.
22. Blum WF, Englaro P, Hanitsch S, Juul A, Hertel NT, Müller J, Skakkebaek NE, Heiman ML, Birkett M, Attanasio AM, Kiess W, Rascher W. Plasma leptin levels in healthy children and adolescents: dependence on body mass index, body fat mass, gender, pubertal stage and testosterone. *J Clin Endocrinol Metab* 1997; 82:2904-2910.
23. Wabitsch M, Blum WF, Mucbe R, Braun M, Hube F, Rascher W, Heinze E, Teller W, Hauner H. Contribution of androgens to the gender difference in leptin production in obese children and adolescents. *J Clin Invest* 1997; 100:808-813.
24. Russ MJ, Ackerman SH, Barakat R, Levy B. Hypogonadotropic hypogonadism and delayed puberty in a man with anorexia nervosa. *Psychosomatics* 1986; 27:737-739.
25. Wesselius CL, Anderson G. A case study of a male with anorexia nervosa and low testosterone levels. *J Clin Psychiatry* 1982; 43:428-429.
26. Ohzeki T, Shiraki K, Igarashi Y, Egi S, Kamoshita S, Kobayashi N. A 25-year-old man with congenital hypothalamic hypogonadism and anorexia nervosa. *Exp Clin Endocrinol* 1991; 97:97-98.