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Case Report

Resolution of Severe Anorexia Nervosa Associated with Gender Dysphoria (Dysphorexia) by Testosterone Therapy in Two Male Transgender Youth

Francisco Javier Martinez-Martin,[1]*Alba Hernandez-Lazaro,[2] Paula Maria Fernandez-Trujillo-Comenge,[2] Ricardo de Leon-Durango,[2] Paula Gonzalez-Diaz,[3] Esther Hernandez-Felipe,[4] Maria del Pino Perez-Garcia,[5] Agnieszka Kuzior[6]

[1]Gender Dysphoria Clinic, Hospital Universitario de Gran Canaria Doctor Negrin, Spain

[2]Endocrinology & Nutrition Dpt, Hospital Universitario de Gran Canaria Doctor Negrin, Spain

[3]Centro de Salud de Arucas (Primary Healthcare Center), Arucas, Gran Canaria, Spain

[4]Centro de Salud de Guanarteme (Primary Healthcare Center), Las Palmas de Gran Canaria, Spain

[5]Facultad de Ciencias de la Salud (School of Medicine), Universidad de Las Palmas de Gran Canaria, Spain

[6]Endocrinology & Nutrition Dpt, Hospital San Roque Meloneras, San Bartolome de Tirajana, Spain

Abstract

Anorexia nervosa is a severe and potentially lethal eating disorder. In transgender youth with severe gender dysphoria, a severe eating disorder (proposed name: dysphorexia), coherent with anorexia nervosa may be triggered by the desire to avoid the cisgender pubertal transition. In these patients, gender-affirming hormone therapy can be extremely effective. We report hereby the cases of two female-to-male transsexual patients with severe gender dysphoria whose anorexia nervosa was related to their pubertal development and who promptly recovered when they started gender-affirming hormone therapy with testosterone, after very limited success with standard psychotherapy and pharmacotherapy for anorexia nervosa. Our patients could not access pubertal suppression due to lack of parental consent in one case and failure to express the conflict in the other. We postulate that avoiding the cisgender pubertal transition with GnRH agonist treatment might also be able to prevent the development of dysphorexia.

Introduction

For many adolescents with gender incongruence, the pubertal physical changes are a cause of intense dysphoria and unbearable suffering.[1] Early treatment (typically beginning in the Tanner pubertal II stage) with GnRH analogues in order to suppress puberty has been shown to improve psychological functioning and physical outcome.[2] Anorexia nervosa is a severe and potentially lethal eating disorder, characterized by a restriction of energy intake relative to requirements leading to an abnormally low body weight; an intense fear of gaining weight and a distorted perception of body shape.[3]

The general cause of anorexia nervosa is unknown,[2] but in the case of adolescent transgender people, a specific issue with the perception of body shape and development is apparent. The restriction of energy intake (often associated with excessive exercise, vomit induction or use of laxatives) may impede or delay the pubertal development of the secondary sex characteristics associated with gender dysphoria, alleviating the suffering of the patient. Although the present American Psychological Association[4] guidelines for practice with transgender and gender nonconforming people do not mention anorexia nervosa, there appears to be a higher prevalence

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***Corresponding author:** Francisco Javier Martinez-Martin, Endocrinologist, Postal Address: Gender Dysphoria Clinic, Hospital Universitario de Gran Canaria Doctor Negrin, Barranco de la Ballena s/n, 35010 Las Palmas de ran Canaria, Canary Islands, Spain

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of documented eating disorder symptoms in transgender youth compared to cisgender youth.[5] Reportedly, transgender youth with eating disorders are at particularly high risk for self-injury and suicidal behaviours.[6,7] In a few of the reported cases,[8,9] the pathological eating behaviour of the patients was explicitly reported as a way to manage gender expression (menstruation cessation, loss of secondary sexual characters). In a report, puberty suppression with a GnRH agonist quickly restored healthy eating habits and normal psychological functioning in two transgender adolescents.[10] There is also a paucity of reports of the effect of gender-affirming therapy in post pubertal patients. In one case report, a patient initially presented as a 16year-old typical female with anorexia nervosa was later revealed as a male transgender subject in whom treatment with testosterone plus bilateral mastectomy alleviated his eating disorder.[11] Another case report describes the partial recovery of a 19-year old female transgender subject with anorexia nervosa after she received suppressive therapy with leuprolide and spironolactone, although she did not receive estrogen therapy.[12] The association between gender dysphoria and eating disorders has been explored in two recent reviews[13,14] but the conscious link between caloric restriction and the avoidance of pubertal cisgender development has very rarely been reported.

We report hereby the cases of two male transgender patients (female-to-male transsexuals) with severe gender dysphoria who did not have access to GnRH agonist therapy at the onset of puberty, and who were diagnosed with severe, life-threatening anorexia nervosa. In both cases, the eating and behavioural disorder was explicitly linked with the rejection of their pubertal physical development, and was promptly resolved when they started gender-affirming hormone therapy with testosterone. We consider that, while this syndrome is congruent with the current diagnostic criteria of anorexia nervosa,[2] has a peculiar etiology (the desire to avoid the cisgender pubertal development and the associated dysphoria) and is amenable to a peculiar treatment (puberty suppression with GnRH agonists or gender-affirming hormone therapy) which would not make sense for the general (i.e. non-transgender) patient with anorexia nervosa; we would like to propose a new denomination for this syndrome, namely dysphorexia.

Case Reports

I. A highly intelligent child, assigned female at birth (with normal karyotype 46XX and no developmental anomalies) had been diagnosed before puberty with Asperger syndrome and had gender incongruence. By age 11 he was at Tanner II puberty stage, whence he experienced intense gender dysphoria centered in the anticipation of menstruation and breast development. He did not start the recommended GnRH agonist treatment at the Tanner II puberty stage by age 11 because parental consent was not obtained. He developed a severe eating disorder diagnosed as anorexia nervosa, with severe caloric restriction and excessive exercise, requiring several admissions in a specialized unit and forced enteral nutrition. By his 14th birthday he was transferred to the attention of our Gender Dysphoria Clinic. He weighed 34kg with a height of 162cm (BMI<13kg/m^2). He had Tanner III axilarche and pubarche, Tanner II thelarche, and had experienced no menarche. His malnutrition was obvious, with emaciation and low plasma albumin (2.2g/dL). Both psychotherapy and pharmacologic therapy had been largely unsuccessful. At the age of 14yr 6 months parental consent for gender-affirming hormonal therapy was obtained, after reviewing with both parents the possibility of a resolution of the eating disorder. The patient began treatment with GnRH agonist and progressive transdermal testosterone treatment up to 60mg/day. One year later his eating habits were normal, his BMI was 19.85kg/m^2 (height 177cm, weight 62.2kg), his FSH, LH and testosterone were in the normal male range and lab tests showed no nutritional deficit except for low vitamin D (22ng/mL), due to his avoidance of outdoor activities; a cholecalciferol supplement has been recommended.

II. A 20year-old youth, assigned female at birth, (also with normal karyotype 46XX and no developmental anomalies) was interned in the Psychiatry Ward of a General Hospital due to extremely severe anorexia nervosa, with BMI<11kg/m^2, extreme emaciation, very low plasma albumin (1.9g/dL) and inability to walk. During the admission, he reported that he identified with the male gender since childhood but had never expressed it socially, not even to his parents. He had experienced extreme dysphoria during pubertal development with normal breast development, menarche at 12years and normal menses until the onset of anorexia nervosa. When offered gender-affirming hormone therapy and bilateral mastectomy (after a reasonable recovery), he accepted enteral feeding via nasogastric tube and was discharged after his BMI approached 16kg/m^2 and had recovered his ability to walk and take care of himself; he was then referred to our Gender Dysphoria Clinic. He started parenteral testosterone cypionate treatment, beginning with 25mg weekly, progressively escalating doses up to 200mg biweekly. In 6months his recovery was obvious, with no nutritional deficit in his lab tests. 14months after discharge his eating habits remain normal, his BMI is 20.4kg/m^2, his FSH, LH and testosterone are in the normal male range and he has no nutritional deficit, but his mastectomy is still pending (unfortunately delayed due to the COVID-19 pandemic).

Conclusion

In transgender patients with severe gender dysphoria, a severe eating disorder (proposed name: dysphorexia, coherent with anorexia nervosa but with a peculiar etiology and therapeutic approach) may be triggered by the desire to avoid the cisgender pubertal transition. In these patients, gender-affirming hormone therapy has been extremely effective, contrasting with the poor result of standard psychotherapy and pharmacologic therapy. It has previously been reported that avoiding the cisgender pubertal transition with GnRH agonist treatment may be able to revert the abnormal behavior pattern of anorexia nervosa[10] and we postulate that implementing this therapy in early puberty (Tanner II stage) might impede the development of eating disorders in transgender youth who experience severe gender dysphoria. Unfortunately, some

youth transgender people cannot access puberty-suppressing therapy for a variety of reasons, such as unawareness, unavailability or high cost; in our cases lack of parental consent (required by law) or failure to express the conflict. Our experience shows that gender-affirming therapy with testosterone may be able to resolve dysphorexia in male transgender youth with severe gender dysphoria, both when the pubertal development has been incomplete due to severe caloric restriction, and well after the pubertal development has been completed.

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Conflicts of interest

Authors declare that there is no conflict of interest.

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