A case report of comorbid spina bifida and anorexia nervosa

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SUMMARY

Young women with physical disabilities are known to have a higher risk of developing any type of eating disorder. Because the physical disabilities of these patients usually include components such as body image disturbances, feelings of lack of control arising from the need to get help from others and excessive attention to maintaining the weight that will allow the desired mobility. Although there is such a relationship between disabling chronic diseases and eating disorders, there are a limited number of publications on this subject in the literature. Spina bifida is one of the most common congenital defects of the central nervous system and occurs in around 1 per 1000 births worldwide. Many physical, medical, cognitive, emotional and psychosocial secondary consequences are observed in spina bifida patients and these consequences create a serious disease burden. Body dissatisfaction is prevalent in patients with spina bifida and body dissatisfaction has been identified as a risk factor for the development of eating disorders. Anorexia nervosa is a serious mental disorder characterized by excessive fear of gaining weight and disturbed body image. During anorexia nervosa, severe weight loss and secondary medical problems that may have a life-threatening impact on the patient may occur. In this case report, the diagnosis and treatment process of anorexia nervosa in a young female patient with spina bifida is discussed in detail.

Key words: Anorexia nervosa, spina bifida, disability

INTRODUCTION

Spina bifida (SB) is a congenital malformation that occurs as a result of failure to close the embryonic neural tube during the fourth week of pregnancy after fertilization (1). Although the prevalence rate varies between countries, it is the most common birth defect of the central nervous system. The clinical manifestation of SB depends on the level of the lesion in the spine and the presence or absence of hydrocephalus (2). Myelomeningocele is the most prevalent and most severe anatomical subtype of SB and accounts for more than 80% of cases. Myelomeningocele is a midline anomaly generally in the lumbosacral region where the skin is deficient and it simply appears ulcerated with an exposed placode comprising primitive neuronal epithelium (3). Patients with myelomeningocele often exhibit motor and sensory neurological deficits below the level of lesion (2). SB is a chronic disease that can require regular care for bladder or bowel problems and cause orthopedic abnormalities. Also intellectual disabilities and difficulties with social skills may be observed in SB. For all these reasons, patients need to use ambulatory aids and independent functioning becomes more challenging (4).

Anorexia nervosa (AN) is a mental disorder characterized by intense fear of gaining weight, disturbed body image, low body weight, severe dietary restriction or weight loss-oriented behaviors. Its lifetime prevalence is around 1% in women and less than 0.5% in men, and it often begins in adolescence. The etiology of AN is complex and includes a combination of genetic, developmental, psychological, familial and socio-cultural factors (5). Increased rates of medical morbidity, mortality and psychiatric comorbidity have been detected in AN. The disorder is often characterized by relapsing or chronic courses in which patients often discontinue their treatment (6).

To date, it has been reported that young women

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with physical disabilities can display inappropriate eating and weight control behaviors on a spectrum ranging from unhealthy diet to eating disorders (7–9). The first report about eating disorders in patients with SB was in a case series published in 1997. It was stated that all cases were premorbidly overweight and developed eating disorders over the years in order to reduce their neurological limitations and increase their mobility (10). In this case report, the development, diagnosis and treatment process of AN in a patient with SB is discussed in detail.

Case history

A 22-year-old female patient with no previous history of psychiatric diagnosis or treatment was referred to the psychiatry outpatient clinic from the gynecology department with the complaint of severe weight loss, impaired body image, restriction of eating, amenorrhea and depressive symptoms. She was single, had a 2-year university degree and was not working in any job. Before applying to our hospital, she was followed up in a state hospital for 3 months with a diagnosis of AN. During this period, she used fluoxetine 40 mg/day and aripiprazole 5 mg/day treatment, but no significant improvement was achieved. The patient, who was 1.48 m tall, weighed 27 kg and had a body mass index (BMI) of 12.3 kg/m2, was referred to our university hospital for hospitalization. She had SB in form of myelomeningocele and was mobilized with an electric wheelchair. She was admitted to the psychiatric inpatient unit for treatment.

During psychiatric interviews, she stated that her orthopedic physician recommended her to lose weight due to increased mobility problems one year ago and her father put intense pressure on her for this reason. She weighed 58 kg and her BMI was determined as 26.8 kg/m2. She was overweight (BMI: 25-29.9 kg/m2) according to BMI based classification. From this date onwards, she severely restricted her food intake and began to hide some foods. She lost approximately 30 kilos in one year, but still felt very overweight. She set her target weight as 25 kg. She also had symptoms such as unhappiness, anhedonia, irritability, intense fear of gaining weight and fatigue.

She is the first child of the family and there is no consanguineous marriage between her parents. Her mother visited a doctor only once during the pregnancy and the patient was born via spontaneous vaginal birth. She was diagnosed with SB after birth and was operated for myelomeningocele (between L2-4) when she was a 46-day-old baby. She had concurrent congenital hip dysplasia and was operated for this reason when she was 6 months old. After this operation she could only walk with support at the age of 6. At the age of 11, Chiari II malformation and non-communicating hydrocephalus were detected in brain imaging. The latest magnetic resonance imaging result is shown in Figure 1. Since the patient remained stable, there was no need for a shunt operation. Her mental development has progressed in line with her peers. She started using a wheelchair at the age of 13 due to increasing orthopedic problems. Mitrofanoff operation (appendicovesicostomy) was performed at the age of 15 due to neurogenic bladder. After the operation, she started to use clean intermittent catheterization and she was defecating by being taken to the toilet at regular intervals with the support of her mother. While she could initially sit in a wheelchair by herself, in recent years she needed to be held for non-wheelchair mobilization. During holdings, her father often gave feedback that she was too heavy and he had difficulty carrying her. After starting to use wheelchair, her depressive symptoms and body dissatisfaction gradually increased. That's why she didn't want to look at her own body in the mirror.

Figure 1: Cerebellum extending into the spinal canal (Chiari II malformation)

She also had chaotic family dynamics. She has two other healthy younger siblings. Her father was hearing impaired and had difficulty controlling his anger. Her mother was mentally exhausted due to both housework and patient's care. Her parents had marital problems and her father sometimes used physical violence against her mother.

The patient believed that her physical limitations due to SB made her a burden to her family and that she was incapable of contributing to anything meaningful. She felt that she had no control over anything in her life and frequently experienced episodes of anger outbursts. She constantly expressed that she was not thin enough and, as a result, persisted in severe food restriction. Body dissatisfaction was contributing to social isolation and her depressive symptoms continued within this vicious cycle. The patient exhibited minimal insight regarding AN and attributed all her problems exclusively to SB.

After admission to the psychiatry inpatient unit, laboratory examinations revealed hypercholesterolemia (Total cholesterol: 216 mg/dL and LDL: 142 mg/dL). Erythrocytes, leukocytes, protein and bacteria were observed in the complete urine analysis. Escherichia coli and klebsiella oxytoca growth was detected in the urine culture. With the recommendation of infectious diseases, ceftriaxone 2 g/day was administered intravenously for 14 days. Sinus rhythm was detected on electrocardiography. The heart rate was 80 beats/min and there were no abnormal waves. The patient was consulted to the nutrition and diet unit. Her diet was adjusted and a supplement containing glutamine was prescribed. Oral vitamin D and iron replacement were administered. Gynecology did not make any additional recommendation other than planning weight gain. The patient was included in a daily physical therapy program for chronic loss of muscle mass and strength. Additionally, home exercises were planned by physiotherapists. Psychoeducation interventions were applied to her family and the parents were referred to the psychiatry outpatient clinic due to the difficulties they experienced. During her 8-week hospitalization, her treatment was arranged as fluoxetine 60 mg/day, olanzapine 5 mg/day, mirtazapine 15 mg/day and clomipramine 50 mg/day. The patient's depressive symptoms decreased, her treatment compliance increased and she was discharged when she weighed 35 kg and her BMI was 15.9 kg/m2. After discharge, she has continued to be followed up at the psychiatry outpatient clinic once every two weeks for supportive psychotherapy and drug treatment. As a result of this three months follow-up, her current weight is 42 kg and her BMI is 19.1 kg/m2 (normal range: 18.5-24.9 kg/m2).

DISCUSSION

Patients with physical disabilities are at high risk for developing unhealthy eating behaviors on a spectrum from obesity to AN (8,11). In a clinical study in which patients with sequelae due to SB and rheumatological diseases were evaluated for eating disorders, the rate of eating disorders in the patients was determined as 8% (8). Considering that eating disorders are observed between 0.5-2% in the normal population, the prevalence of eating disorders in physically disabled individuals has increased significantly. However, prevalence of disordered eating is unknown in adolescents and young adults with SB. In a national survey study including adolescents and young adults, both males and females with SB exhibited substantially higher purging and restricting behaviours compared to healthy controls (11). Eating disorders are also more common in women than in men with physical disabilities (9). In a series of 5 case reports with SB and eating disorders, AN was detected in 3 of the cases and an eating disorder not otherwise specified was detected in the other two. It was reported that all cases were initially overweight and subsequently lost weight to reduce physical limitations (10). Eating disorders occur at a later age in people with physical disabilities and chronic diseases. This may be a reflection of maturity lag. In these patients, the transition to adulthood is more related to the age to achieve developmental milestones such as movement away from parent, development of intimate relationships, financial and economic independence etc. rather than chronological age (8). Our patient has similar characteristics to the cases in the literature in that she was initially overweight, started to lose weight after her physical limitations increased, was dependent on her parents' care for her daily life and her disease started later than expected at the age of 21.

A disturbance of body image has a core role in the psychopathology of AN. Body image is a multidimensional concept, encompassing elements of body perception (estimation of physical aspects of the body and its functions) and attitude (body valuation and esteem) (12). The prevalence of body dissatisfaction in young women with physical disabilities is approximately %35 (13) but sometimes this rate can exceed above 60% (8). It has been reported that body image in people with physical disabilities is affected by factors such as immobility level, visual and tactile stimuli, neurodevelopmental disorders, enmeshed family relationships, others' opinions and recommendations of healthcare professionals.

Patients with mobility-related disability engaged in eating disorder practices in order to compensate for their disability and avoid being double handicapped (9). Our patient stated that her dissatisfaction with her body and thoughts of inadequacy and failure increased, especially after she started using a wheelchair. Conditions that prevent even driving are risk factors for eating disorders in physically disabled patients (8). The fact that the patient has been using a wheelchair for a long time and has been mobilized by being carried on the lap when not in a wheelchair in recent years may be an important risk factor for the development of body dissatisfaction and AN.

Mental body representations consist of the integration of much information arising from the shape and size of body parts, their position in space and their relationship with other body parts. These representations are constructed from and reciprocally influenced by visual and tactile inputs (14,15). Moreover, certain aspects of body representations may not only be influenced by bottom-up sensory input, but also by top-down cognitive, semantic and affective elements (14). Our patient had sensory loss bilaterally in her legs due to myelomeningocele. She avoided looking in the mirror because of her body dissatisfaction. She was trying to understand the thickness of her legs and waist by measuring them with her hands. Despite her emaciated appearance, she experienced her body as too fat. It has been observed that patients with AN visualize their bodies less precisely and overestimated distances between tactile stimuli on different body parts compared to healthy controls (14). When somatosensory inputs do not provide sufficient bottom-up information, top-down information may play a more important role in shaping body image. This disturbance in body image could be a key factor in the development and maintenance of AN in our case.

Congenital abnormalities of hindbrain structures with bowing and elongation of the medulla oblongata and pons, and descent of the cerebellar vermis below the craniocervical junction occur in Chiari II malformation. Chiari II malformation almost always accompanies myelomeningocele (16). Patients with SB may have less visible executive dysfunction such as solving novel problems, modifying behaviour in the light of new information, generating strategies etc. as well as severe mental retardation (17,18). Cognitive impairment in SB-Chiari II complex is affected by the severity of the hydrocephalus and shunt-related complications (19,20). Our patient has Chiari II malformation and non-communicating hydrocephalus. The patient's condition has been stable for many years and there is no need for a shunt. However, subtle cognitive impairments that may occur in the patient due to this malformation and hydrocephalus may cause the disease to persist and slow recovery in treatment. Sensory input originating from the gastrointestinal system enters the brain by being transmitted to the nucleus tractus solitary through the vagal nerve (21). It has been shown that interoceptive sensitivity in certain visceral organs is reduced in AN compared to healthy controls (22). In our case, the anatomical structures that regulate appetite at the brainstem may have been affected due to Chiari II malformation and the perception of sensory stimuli arising from visceral organs may have been impaired due to myelomeningocele at the L2-4 level. These conditions may facilitate the development of AN in our case.

In patients with physical disabilities and developing eating disorders, body image is influenced by more in line with the opinions of other people rather than the media. Family members, peers and medical/social care professionals come first especially with their criticisms and suggestions about weight among these people (9). Considering that people with physical disabilities are dependent on their

family members and have more frequent contact with healthcare professionals than usual, their feedback on body perception becomes more important. Also eating disorders often involve enmeshed family dynamics that are excessively intrusive and have uncertain boundaries (8,9,23). The patient's physical dependence on her family, her family's inability to adequately evaluate the effect of feedback and the doctor's directive approach to losing weight may have contributed to the development of AN in our patient. We tried to optimize the symbiotic interaction between the patient and her parents during her hospitalization. We helped her parents to understand factors that may play a role in the development of AN. We determined the treatment goal with the patient to reach and maintain a healthy weight rather than constantly losing weight.

Relapse rates in AN range from 30% to 50%. Predictors of relapse in AN within categories such as age and sex, symptoms and behaviors, AN subtype and duration, weight or weight change, comorbidity and personality were evaluated in a recent systematic review. Solely pre-treatment depression and lower post-treatment BMI were found to be statistically significantly associated with AN relapse (24). In our case, the presence of pre-treatment depression, a low BMI of 15.9 kg/m^2 at discharge and 19.1 kg/m^2 during follow-up can be considered risk factors for relapse. Somatosensory impairments associated with SB can complicate the treatment of body image disturbances. Furthermore, due to deformities and disabilities related to SB, complete resolution of body dissatisfaction appears unlikely. Considering these clinical features, the prognosis for this case is anticipated not to be very favorable and the treatment process is expected to be particularly challenging.

Nowadays, 75% of patients with SB reach young adulthood and approximately one-third of the patients are hospitalized for preventable secondary conditions (25). More studies are needed to identify different risk factors for eating disorders in physically disabled patients and to apply more effective strategies in their treatment.

Informed Consent: Written informed consent was obtained from the patient to publish this case report.

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