

An Adolescent With Anorexia Nervosa and Gastrointestinal Stromal Tumors

Gladys J. Frankel, Ph.D.

Katherine A. Halmi, M.D.

The literature often describes medical complications that are a result of the malnutrition and weight loss that define anorexia nervosa. The comorbidity of anorexia nervosa with other independent medical disorders associated with weight loss is unusual. Case reports have described anorexia nervosa coincident with, for example, lymphocytic leukemia (1), chronic pancreatitis (2), celiac disease (3), and cerebral meningioma (4). In the present report we describe the case of a young man who met diagnostic criteria for anorexia nervosa and gastrointestinal stromal tumors. This case presentation emphasizes the importance of making a diagnosis based on positive criteria. Distinguishing features of the two disorders are highlighted in the process of differential diagnosis. Treatment decisions and relapse issues are presented for both conditions. The patient is given the name Alex in this report to protect his identity.

Case Presentation

Patient History

Alex, a 16-year-old single Caucasian boy, was mildly overweight from the first few months of life throughout his childhood. According to his parents, his pediatrician was never concerned and stated, "He'll grow out of it." Alex was a talented athlete, active in many sports, and his overweight condition did not impede him until he reached 13 years of age. Then at 5'8", he had a maximum weight of 245 lb. At this time, his baseball coach advised him to lose weight and frequently benched him because of his obesity. In response to the additional stress of teasing from his peers, Alex began to diet. Initially he restricted intake of foods that had high fat content, and then he began to compulsively count calories in all foods. He gradually restricted his diet to liquid egg substitute and increased his exercising rituals. After 7–8 months, at age 14, he had lost 100 lb. Alex continued to severely restrict his food intake and overexercise over the following year and a half.

In 1998, when Alex was 16 years old, he had symptoms of fainting and bleeding, with considerable blood loss from a bleeding gastric ulcer. Endoscopy examination showed a tumor in the lower part of his stomach. Subsequent surgery that day removed a gastrointestinal stromal tumor the size of a "golf ball." The surgeon spec-

ulated that the tumor had grown to that size over 1 year. Alex was postoperatively clear of any observed malignancies for 3 years.

Because Alex was continuing to restrict his food intake, at the insistence of his parents he began therapy with a nutritionist and a psychiatrist in December 1999. A month later, in January 2000, he was abusing laxatives, running 3 to 4 miles, and doing 2,000 sit-ups each day. At this time his weight was 100 lb. He was seen by a pediatrician, who was not concerned and did not require that he seek further treatment, such as hospitalization, for his eating disorder. His parents, however, were very concerned and thought he could benefit from more specialized treatment. Hence, they sought another physician, who helped them with the appropriate referral to our eating disorder unit at the New York Presbyterian Hospital—Westchester Division.

At admission in January 2000, Alex was 5'8" tall and weighed 101 lb. The treatment focused on weight gain and breaking his cycle of disordered eating behaviors with cognitive behavior techniques. His discharge weight was 130 lb; the recommended target weight was 142 lb. The treatment team made specific recommendations for ongoing caloric intake, structured eating times, and weight goals. Rehospitalization was recommended if Alex lost more than 5 lb. After discharge, Alex did not feel that any of the adults in his life were enforcing the hospital's guidelines, so he decided to do what he desired. He minimized the importance of eating and was not successful at gaining weight. Five months later,

Alex was readmitted a second time for his eating disorder, weighing 10 lb less than his previous discharge weight. During this hospitalization he refused to achieve a weight greater than 130 lb, although his ideal target weight was 142 lb. He left the hospital against medical advice. An outpatient multidisciplinary therapy program was initiated for Alex. This included psychotherapy with a psychiatrist, counseling with a nutritionist, and family therapy with Alex's parents. In January 2001 the nutritionist changed jobs, and Alex did not seek another referral. He attended his therapy sessions sporadically, and his parents were frustrated in their attempts to be part of his treatment program. Alex completed high school during this year and was awarded a full scholarship to a local university.

In August 2001, Alex had blood tests, which showed abnormal elevation of liver enzyme levels. The computerized tomography (CT) scan raised suspicions about metastatic disease and showed three visible tumors. Subsequent liver biopsies at a major cancer research hospital confirmed reoccurrence of his primary cancer. Alex was enrolled in an experimental study with imatinib me-

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sylate, a specific inhibitor of the tyrosine kinase produced by the tumors' mutant c-kit proto-oncogene (5). After administration of imatinib mesylate for a few months, the CT scan demonstrated a slight improvement in the liver metastases. Alex did not enroll in college in September 2001. His father believed Alex refused to attend college because he wanted to continue to engage in his eating disorder behaviors. At this time his local psychiatrist prescribed olanzapine in an attempt to help reduce his eating-related preoccupations and purging.

In January 2002, there was a transient edema in Alex's lower extremities coinciding with more severe food-restricting behavior. He was hospitalized locally for severe weight loss, severe food restriction, and laxative abuse. At this time the CT scan demonstrated a decrease in the previous metastases and no new lesions. His parents wished Alex to be readmitted to our eating disorder unit; however, there were two problems. One, Alex was 19 and refusing hospitalization, and his parents did not wish to have him committed; second, the imatinib mesylate was an experimental medication, not yet approved by the Food and Drug Administration (FDA), and Alex's parents were concerned that Alex would not be able to continue taking the medication at a different hospital.

In February 2002, Alex had recurrent hospitalizations at his local general hospital with progressive weight loss that disqualified him for the chemotherapy protocol, and the imatinib mesylate was discontinued. Alex refused the recommendation for readmission to our eating disorder unit, and repeated attempts at multidisciplinary outpatient therapy failed as he progressively lost weight. In March 2002, after achieving some weight gain at the local hospital, Alex signed himself out against medical advice only to be readmitted after 3 days at home because of an immediate relapse with severe laxative abuse. At this time he acknowledged his need for help. A repeated CT scan demonstrated new metastases to the liver. Chemotherapy with imatinib mesylate was resumed as the medication was fully approved by the FDA and he was no longer in an experimental protocol. The local general hospital treatment team facilitated admission to our eating disorder unit with the agreement of both Alex and his parents.

Alex was admitted to the eating disorder unit for the third time in April 2002. At this time his height was 5'7.25", lower than his previous height of 5'8". His weight was 107.6 lb, which represented a loss of 22.4 lb from his discharge weight of 130 lb 1.5 years before. Alex met diagnostic criteria for anorexia nervosa, binge/purge type. Alex severely restricted his food intake, eating only two meals a day with the same content of egg substitute and "lite" bread. Between the meals he would purchase \$40 worth of high-calorie snack foods, which he would chew and then spit out. He drank 10 to 15 cups of coffee a day to curb his appetite. His parents informed the treatment team that there were large garbage bags of masticated food hidden throughout Alex's apartment. His parents also stated that he consumed several pounds of butter or frozen dessert topping in the course of a week. The latter, Alex informed us when confronted, facilitated regurgitation. He continued consuming one box of laxatives a day, a habit that had been ongoing for several years. He often stole laxatives and spoke of being afraid to stop using them, fearing that he would be unable to move his bowels and would live in a continuous

state of constipation. He learned that an excessive dose of magnesium hydroxide antacids would cause diarrhea, and he would place the antacids in an empty can of nuts so he could pretend that he was eating nuts. Alex dreamed of becoming a gourmet chef and was very facile with nutritional information, citing calorie counts for nearly every food on grocery shelves. His conversations with his parents consisted solely of food-related topics.

Family History

There was no family history of mental illness or eating disorders. An older sibling who lived outside the family residence and a younger half-sibling were reported as being without problems. Alex's parents had divorced when he was 6 years old, and his mother had been remarried for 12 years. His mother and step-father and his biological father had difficulty controlling Alex and setting limits. Recently he had manipulated them by requesting to live with an elderly neighbor to help with her care. When the parents consented, in this new setting he engaged in his disturbed eating behaviors with no inhibitions.

Course of Treatment

In the third hospitalization for the eating disorder, the treatment focused on weight gain, changing restrictive eating habits, breaking binge/purge cycles, promoting a commitment to weight maintenance, and learning cognitive behavior techniques to counteract dysfunctional thoughts and behaviors. Alex benefited from the multimodal treatment of a highly structured inpatient unit where he was supervised and could not engage in his disordered eating behaviors. His total caloric intake was given as food supplemented by liquid formula at regular intervals, and the amounts were increased every few days. Alex now understood the importance of maintaining an adequate weight, since he realized that treatment with imatinib mesylate had to be discontinued if he was below 100 lb, and when that occurred the metastases would recur. Although he was motivated to be cooperative, Alex did not wish to discuss his cancer. His nutritional education strengthened his perception that maintaining a body at an appropriate weight developed a nutritionally strong body, which was helpful in fighting cancer. A self-perception of being strong can be beneficial for cancer patients according to Spiegel et al. (6), and this approach was reinforced by the therapy staff.

Cognitive behavior exercises were used daily to help Alex challenge his intrusive thoughts that weight gain was synonymous with being fat. A cost-benefit analysis reinforced his knowledge of the consequences of his disordered eating behaviors and helped motivate him to improve his habits. Cognitive restructuring exercises, reviewing the thought that gaining weight equals being fat and identifying facts to support this belief and facts disproving the belief, helped him develop a logical thinking process to handle his distorted beliefs. Problem-solving exercises were used to handle his urges to restrict food intake and to spit food out. They also identified more effective coping skills. A very important and poignant issue for Alex was being in control. Among the many interactions that played out during his treatment, Alex was angry when he was not in control. He was particularly agitated by the fact that he could not control what he was given to eat, when he was to eat, or when he was to be

discharged. He eventually came to realize that his choice to cooperate and eat was far better for him than his prior choices of restricting food intake. As his medical condition improved, he was given the choice of his snacks, which challenged him to control his urges to binge. Some of his early food choices required redirection, but over time he was able to make appropriate choices and feel more confident.

Another theme for Alex was the perception of being ignored. Patients are encouraged to make requests, and whenever Alex made a request and the request was not granted, he would bitterly complain that he was not being listened to. At the end of treatment he was provided with an opportunity to choose his own foods, so that this opportunity would reinforce his sense of having a voice and being successful. Although the staff was divided about this decision, research by Spiegel et al. (6) showed that cancer patients who had opportunities to express their feelings did better than patients who did not participate in groups to express their emotions. Spiegel and colleagues also commented anecdotally that cancer patients who seemed to be the most annoying and demanding to their doctors had better survival rates.

To help reduce Alex's obsessive thoughts about calories and eating, olanzapine, 5 mg/day, was given in an attempt to reduce these obsessive and distorted thoughts. He was also given sertraline, 150 mg/day, for mild depressive symptoms and bulimic symptoms. At the time of discharge, Alex thought the medications were helpful in allowing him to concentrate on enjoying his food without being obsessively concerned about the calories or fat content. It was recommended that he continue to take these medications during his intensive outpatient treatment program.

During the course of hospitalization, family therapy was conducted by telephone with Alex's parents since they were geographically far away. His parents participated in person in a final session at the time Alex was discharged.

Alex was discharged from the eating disorder unit weighing 135 lb, 95% of his target weight of 142 lb. His mood and thinking had greatly improved. The recommendation to participate in a transitional day therapy program for eating disorders and a support group for cancer patients met with agreement from Alex and his parents. He was encouraged to take a responsible role in the care of the family dog, to whom he was attached, and to participate in activities with his father to support his commitment in following through with discharge plans. Research on pet therapy (7) has demonstrated that providing care to a pet is similar to promoting care of one's self.

Discussion

Anorexia nervosa is a disorder characterized by weight loss, fear of gaining weight, and a denial of the consequences of the weight-losing behaviors. The initial food avoidance becomes an all-consuming food phobia, which is necessary to maintain the weight loss. This in turn can serve a positive function in the patient's life, providing escape from aversive issues and distressing life events. The disorder becomes highly reinforcing, and the prospect of relinquishing the anorectic behavior patterns is often ter-

rifying to the patient. Alex developed anorexia nervosa at age 14, when he responded to the demands of his coach to lose weight and the teasing of his peers by engaging in severe dieting behavior, which soon consumed his life and all of his attention. Dieting is the most common risk factor for developing an eating disorder (8, 9). This case is an example of the impact that coaches can have on their athletes and the need for educating coaches about healthy eating behaviors. Later, anorexia nervosa also served as a distracter for Alex from the thoughts of the unpredictable gastrointestinal stromal tumors and the metastases.

Gastrointestinal stromal tumors are neoplasms arising from connective tissue elements of the gastrointestinal wall and are a very rare cancer. Clinical findings are gastrointestinal bleeding, abdominal pain, and weight loss (10). This cancer is commonly diagnosed in patients between the ages of 30 and 60 years, is somewhat more common in women, and affects approximately 5,000 people each year. Because of the unpredictable behavior of gastrointestinal stromal tumors and the potential for metastases, patients must regularly be checked and followed indefinitely. Age, tumor size, depth, and location have been found to be predictors of metastases (11, 12), while survival rates of 41% to 83% over 5 years have been reported (11, 13).

For Alex, it is unclear whether the initial 100-lb weight loss was achieved solely by his highly restrictive diet or was abetted by his then-undetected gastrointestinal stromal tumor. Many such tumors are asymptomatic when smaller or have vague nonlocalizing abdominal symptoms. As the tumors become a large mass lesion, they can create an experience of early satiety. The toxicity from the tumor could also have contributed to early satiety. It would be difficult to make a specific attribution since Alex also had strongly held anorectic convictions that directed his behavior. Thus, both processes most likely contributed to the weight loss (14).

The presence of comorbid diagnoses in Alex illustrates the sensitivity and awareness required of physicians to identify eating disorders and distinguish them from other disease processes. Without treatment targeting the anorexia nervosa, treatment of the gastrointestinal stromal tumors alone would have been insufficient to optimize this patient's health. It was imperative to review the presence of characteristic laboratory evidence and the absence or presence of the characteristic psychopathology of anorexia nervosa. Ramsey et al. (15) reported the cases of four men infected with HIV-I whose illnesses exacerbated the symptoms of their eating disorders. In these cases, the comorbidity could have resulted in misclassification as AIDS wasting syndrome, leading to inappropriate treatment. In contrast, there have been a number of cases (16) in which Crohn's disease was mistakenly diagnosed as anorexia nervosa.

The presence of a medical illness providing motivation for a patient to control disordered eating behavior was described by Morgan and Lacey (17) in a woman with HIV-I seropositivity. This diagnosis gave her the impetus to

change her eating behavior. For Alex, his gastrointestinal stromal tumors with metastases contributed to his commitment to weight maintenance, which we hope will increase his lifespan and quality of life. At 1-month follow-up, there were no new tumors and the ones that had been identified were reduced in size. Alex was eating adequate amounts of food, maintaining a good spirit, and enjoying a much-improved relationship with his father.

Cognitive behavior techniques were used in treating both Alex's core anorexia psychopathology and in helping him to deal emotionally with the cancer. On one occasion, when he refused to take imatinib mesylate because he was angry about a later discharge date, Alex participated in a behavioral analysis exercise. This identified how his refusal was designed to express his anger toward his doctors and the staff but was ineffective in that goal and only hurt himself. He then realized his need to use more effective and appropriate coping skills to express his anger. The analysis also showed how he used food restriction and his eating disorder to attack other people. The use of more effective verbal coping skills was reviewed and reinforced. Patients with eating disorders commonly use their bodies (restrict food intake) to express their conflicts because they feel inadequate or insecure in expressing them verbally. Problem-solving techniques were used to focus on issues of assertiveness, expressing one's voice, and the definition of what embodies productive versus ineffective control. The identification of productive control efforts, such as weight maintenance and effective verbal self-expression, were underscored in contrast to the concept of restricting and not verbally expressing oneself, which represent ineffective control. Nutritional education was directed toward a commitment to weight maintenance, reinforcing the benefits of a strong body to fight disease, and the concept that an improved weight equals longer survival. Improved social interaction skills were further emphasized in the context of group therapy. Thus, the cognitive behavior techniques were used for a dual function, to help the patient cope with his cancer as well as overcome the core anorexia psychopathology.

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