Intertrochanteric fracture with low-energy trauma in a young woman with anorexia nervosa
A case report

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Abstract
Rationale: Anorexia nervosa is a chronic psychiatric disease defined by severe weight loss, due to fear of obesity, and self-imposed semi-starvation. Of the many complications following anorexia nervosa, low bone mineral density (BMD) is a significant risk factor for fractures. Anorexia nervosa is associated with higher risk of incident fracture in females across all age groups, and in males >40 years old. Sites at highest risk of fracture include the hip/femur and pelvis in females, and vertebrae in males with anorexia nervosa.

Patient concerns: A 29-year-old woman known to have suffered from anorexia nervosa 15 years ago visited the emergency department due to right hip pain after falling while getting out of a taxi. During the period of anorexia nervosa, she had a body mass index (BMI) of 14.06 kg/m² (weight, 36 kg; height, 1.60 m) and suffered from amenorrhea. At the time of presentation, she had a BMI of 19.53 kg/m² (weight, 50 kg; height, 1.60 m) and had regular menstrual periods, indicating clinical recovery from anorexia nervosa.

Diagnoses: Plain radiography, computed tomography, and bone scintigraphy revealed AO 31-A2.2 type right hip proximal femur intertrochanteric fracture. The BMD showed a T score of −3.9 in the hip and −3.6 at the lumbar level, indicating severe osteoporosis.

Interventions: Osteosynthesis was performed with proximal femoral nail antirotation (PFNA) and wiring.

Outcomes: There were no specific symptoms, such as trauma or infection, during postoperative rehabilitation and postoperative management, and she was discharged after 2 weeks. After 1 year of follow-up at our outpatient clinic, she had no complications.

Lessons: Here, we describe an unusual case of unilateral femoral intertrochanteric fracture that occurred after clinical recovery from anorexia nervosa. This case indicated that the risk of fracture remains even after recovery of BMI. We propose that women who have clinically recovered from anorexia nervosa should be advised to undergo annual osteodensitometric analyses after consulting with specialists in other areas (psychiatry, endocrinology, eating disorders).

Abbreviations: BMD = bone mineral density, BMI = body mass index, MAT = marrow adipose tissue, PFNA = proximal femoral nail antirotation.

Keywords: anorexia nervosa, femoral intertrochanteric fracture, osteoporosis

1. Introduction

Anorexia nervosa is a chronic psychiatric disease associated with severe weight loss, due to fear of obesity, and self-imposed semi-starvation.[1] The definition of anorexia nervosa, as specified in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition, includes body mass index (BMI) <18 kg/m², an intense fear of gaining weight or becoming overweight, disturbance in perception of one’s own body weight or shape, an undue influence of body weight or shape on self-evaluation, and/or persistent lack of recognition of the seriousness of the current low body weight.[2] Many deleterious consequences of anorexia nervosa can be reversed by weight restoration and resumption of normal eating behaviors. However, certain complications, such as low bone mineral density (BMD), can persist for decades after disease resolution[3] and cause lifelong debilitation.

Low BMD may confer a significant risk of fractures.[4] An estimated 50% of women with anorexia nervosa, occurring at both trabecular and cortical sites, have BMD values more than 1 standard deviation (SD) below the mean of a population of young, healthy women, and an additional 35% have bone density values more than 2.5SD below the mean.[6] Vestergaard et al[4] noted that a higher risk of hip fracture in sufferers of anorexia nervosa, including females across all age groups and males >40 years old. The mechanism underlying the change in
bone homeostasis is known to be related to modification of body composition and hormonal changes. With regard to body composition changes, despite low peripheral and visceral fat stores, bone marrow adipose tissue (MAT) inversely increases in anorexia nervosa.[7] Previous studies have demonstrated the clinical significance of elevated levels of MAT, and its relationship to bone density and bone strength. In terms of hormonal changes, hypothalamic amenorrhea is a frequent symptom in females with anorexia nervosa. Hypothalamic amenorrhea results in low androgen levels, in addition to a low estrogen level, which is critical for preserving bone density. Therefore, bone resorption is decreased by the osteoclastic inhibition produced by estrogen.[8] The sites at highest fracture risk include the hip/femur and pelvis in females, and vertebrae in males with anorexia nervosa.[9]

Bardone-Cone et al proposed a definition of clinical recovery from anorexia nervosa in women as a BMI of at least 18.5, with no binge eating, purging, or fasting in the prior 3 months.[10] Weight gain and menstrual recovery are associated with stabilization of BMD. However, complete recovery does not occur, and residual deficits often persist.[11] Therefore, it is obvious that the risk of fracture persists even after clinical recovery from anorexia nervosa.

Here, we report a patient who presented with unilateral femoral intertrochanteric fracture following recovery from anorexia nervosa, with a normal BMI and regular menstrual period.

2. Case description

This case report was approved by the Institutional Review Board of Soonchunhyang University Hospital (IRB No. 2019-02-013). The patient gave written informed consent for publication of this report and the accompanying images.

A 29-year-old woman visited our emergency department due to pain in the right hip after falling while getting out of a taxi. She was known to have suffered from anorexia nervosa with amenorrhea 15 years earlier and had recovered 13 years before presentation. During the period of anorexia nervosa, her weight and height were 36 kg and 1.60 m, respectively, with a BMI of 14.06 kg/m². At that time, she had ingested nothing but water every day for the first 6 months and her weight had decreased from 53 to 36 kg. In addition, she experienced amenorrhea. For the next 18 months, she ate only once a week and experienced irregular menstruation. After this time, she began eating regularly and gained weight gradually. At the time of presentation, she weighed 50 kg with a BMI of 19.53 kg/m², and had regular menstrual periods; this confirmed clinical recovery from anorexia nervosa. The patient had a smoking history of 10 pack-years and consumed alcohol three times per week. Plain radiographs revealed AO 31-A2.2 type right hip proximal femur intertrochanteric fracture (Fig. 1), and computed tomography confirmed intertrochanteric fracture, as seen on plain radiographs (Fig. 2). Bone scintigraphy revealed increased uptake in the right femoral intertrochanteric region suggesting fractures, and there were no other specific findings (Fig. 3). The serum calcium level was 9.1 mg/dL (normal range: 8.0–10.5 mg/dL) and the serum phosphorus level was 3.2 mg/dL (normal range: 2.5–5.5 mg/dL). At the time of presentation, dual-energy X-ray absorptiometry (DXA) was performed. Based on the patient’s age, the T score was −3.9 in the hip and −3.6 at the lumbar level, corresponding to severe osteoporosis.[12,13] Endocrinological studies, including follicle stimulating hormone (FSH), luteinizing hormone (LH), thyroid stimulating hormone (TSH), adrenocorticotropic hormone (ACTH), and growth hormone (GH), were performed but there were no abnormal findings. At 6 days after the injury, osteosynthesis was performed with proximal femoral nail antitrotation (PFNA; Synthes, Paoli, Switzerland) and wiring (Fig. 4). Six

Figure 1. Radiographs of the right hip demonstrating femoral intertrochanteric fracture.
Figure 2. Computed tomography of the hip demonstrating femoral intertrochanteric fracture.

Figure 3. Bone scintigraphy showing mild increased uptake in the right femoral intertrochanteric region.
weeks later, the patient returned to everyday life and reported mild pain at the operation site. Plain radiography at the 6-month follow-up showed complete union of the intertrochanteric fracture (Fig. 5), and gradual advancement to full activity was achieved. She had no complications or recurrence of symptoms at the 1-year follow-up. However, on DXA scan, the T score showed values of −3.4 in the hip and −4.2 at the lumbar level, indicating that she had not fully recovered from osteoporosis.

3. Discussion
To our knowledge, this is the first report of a case of femoral intertrochanteric fracture in a young woman following recovery from anorexia nervosa. At the time of presentation, our patient’s BMI had returned to normal (19.53 kg/m²) and she also had menstrual periods. However, analysis of BMD at the time of presentation showed T scores of −3.0 in the hip and −3.6 at the lumbar level, indicating that the patient still had osteoporosis.
despite clinical recovery from anorexia nervosa. The risk of femoral fracture increases exponentially with age, corresponding with the fact that age is one of the risk factors of hip fracture.[14] However, fractures at the hip are generally considered related to osteoporosis, so a risk of fracture may exist in young patients with low BMD.[15] The decrease in BMD associated with anorexia nervosa involves both trabecular and cortical bone, with the microarchitecture characterized by increased cortical porosity and decreased trabecular thickness.[16] The increased fracture risk after diagnosis suggests that the anorectic state does permanent damage to the skeleton, which may be later exacerbated by the age-related decline in BMD. The first possibility was supported by Herzog et al.[17] who reported reduced BMD even many years after the onset of anorexia nervosa. On the other hand, the anorectic state may not have been successfully reversed, leading to a decrease in bone mass and thus biomechanical competence. This possibility was underlined by studies indicating that BMD recovery was related to disease outcome[17] and a high relapse rate.[18] The low BMD and increased fracture risk at this early stage of life may have long-term consequences.

To improve bone density, the best strategy includes maintaining normal weight and menstrual function. However, complete restoration of bone density to a normal level does not usually occur. Therefore, all patients who have clinically recovered from anorexia nervosa should undergo regular measurement of BMD with DXA. If the BMD does not return to normal after clinical recovery from anorexia nervosa, other therapeutic options to treat low bone density should be discussed. According to the general guidelines, treatment options for low bone density include calcium and vitamin D consumption, hormonal replacement, and pharmacological therapies (denosumab, teriparatide, and/or bisphosphonates).[11] We recommended the above medications to the patient, but she refused due to cost concerns and took only calcium.

Weight-bearing exercises and physical activity are generally recommended in noneating disorder patients with low BMD.[19,20] Mechanical loading has osteogenic properties, positively affects the bone geometry, and leads to increased bone mineral accrual in young patients.[21] Physical activity can also reduce the risk of falls, which in turn decreases the risk of fragility fractures.[22] However, in patients with anorexia nervosa, recommendations are limited and study results vary based on the severity of illness and type of mechanical loading.[23]

Rigotti et al.[24] reported an approximately seven times higher risk of fractures in women with anorexia nervosa compared to an age-matched control population. Vestergaard et al.[9] reported that an increased risk of fractures persists for many years after diagnosis of anorexia nervosa, indicating permanent skeletal damage. According to the literature and the present case report, women with anorexia nervosa should recognize that they are at high risk of fractures and should take additional care in everyday life.

4. Conclusion
We described an unusual case of unilateral femoral intertrochanteric fracture that occurred after clinical recovery from anorexia nervosa. This case showed that the risk of fracture remains even after recovery of BMI after anorexia nervosa. We propose that, after consulting with specialists in other fields (psychiatry, endocrinology, eating disorders), women who have clinically recovered from anorexia nervosa should be advised to undergo annual osteodensitometric analyses and routine pharmacological therapies, to increase or maintain bone density level and thus minimize the risk of hip fracture.

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