

## CASE REPORT

# Management of ischiopubic stress fracture in patients with anorexia nervosa and excessive compulsive exercising

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### SUMMARY

This case report describes a 28-year-old non-athlete female patient with anorexia nervosa who was diagnosed with an ischiopubic ramus stress fracture and treated successfully as an inpatient with a cognitive behaviour-based therapy. The patient's clinical picture, diagnosis and treatment are described, and a brief review of the relevant literature is included. The importance of this case report stems from the rarity of descriptions of this kind of injury in such patients, despite their inherent risk, and the originality of the treatment applied. This, in addition to the usual approach to medical management, exploited specific cognitive and behavioural procedures and strategies to address the patient's excessive compulsive exercising, promoting rest and movement avoidance in order to allow the fracture to heal, while simultaneously addressing the underlying psychopathology.

### BACKGROUND

Stress fractures are a very common phenomenon in sports medicine, and are thought to be caused by continuous and repetitive pulling of the muscles on the bone.<sup>1</sup> Stress fractures can be divided into two types, according to the condition of the bone: fatigue fractures, which occur when bone status is normal; and insufficiency fractures, which occur in weakened bone.<sup>2</sup> Several studies have identified specific clinical risk factors for stress fractures, including low serum levels of 25-hydroxyvitamin D (25OHD),<sup>3</sup> and low mineral content and density in the hip bone,<sup>4</sup> as well as iron deficiency,<sup>5</sup> poor physical fitness,<sup>4</sup> and amenorrhoea.<sup>6</sup> Most stress fractures occur in the tibia, fibula and metatarsal bone<sup>7</sup>; stress fractures of the pubic bone are uncommon, and only few descriptions have been reported. The low risk generally ascribed to pubic fractures increases in long-distance runners and female military recruits, in whom the greatest incidence is reported.<sup>8</sup>

Afflicted individuals usually report the insidious onset of pain over the groin, perineal region, buttock or thigh.<sup>9</sup> Clinical diagnosis is, however, difficult, as these symptoms are frequently confused with muscle injuries such as adductor strain, lumbar disc disease or even referred pain from the gastrointestinal or genitourinary tract.<sup>9</sup> Underweight patients with anorexia nervosa (AN) present an increased risk of stress fractures due to the associated low bone mineral density, which compromises bone structure and strength.<sup>10 11</sup> The reduction of lean and fat masses associated with hormonal alterations secondary to

malnutrition also contribute to impaired bone metabolism in AN.<sup>11</sup> Excessive compulsive exercising,<sup>12</sup> alongside micronutrient and vitamin deficiencies and poor physical fitness,<sup>13</sup> are frequently observed in AN and may further increase the risk of stress fractures, although such cases have seldom been described in the literature. In eating disorder patients, excessive and compulsive exercising has been associated with several distinct features, including elevated eating disorder psychopathology and dietary restraint,<sup>14</sup> higher general psychopathology (particularly anxiety),<sup>15</sup> and obsessive-compulsive symptoms,<sup>16</sup> as well as specific personality features (ie, higher levels of perfectionism, persistence and lower novelty-seeking scores),<sup>17</sup> lower minimum body-mass index (BMI), and younger age.<sup>17</sup> It is also known to obstruct weight restoration in underweight eating disorder patients, and is a predictor of poor treatment outcome.<sup>18</sup>

The only treatment for pubic stress fracture is relative rest and avoidance of physical activity.<sup>9</sup> Crutches may be supplied until ambulatory pain abates, and it may take upwards of 4–8 weeks for complete rehabilitation with optimum healing and return to activity.<sup>9</sup> These procedures should be associated with a treatment aimed to optimise bone density in patients with AN. The most effective strategy to improve bone density in AN is normalisation of weight and restoration of menstrual function.<sup>19</sup> However, recovery from AN does not necessarily ensure the resolution of osteopaenia.<sup>20</sup> What is more, other treatment strategies aimed at improving bone density in patients with AN, such as calcium and vitamin D supplements<sup>21</sup> and the combined oestrogen–progesterone pill,<sup>22</sup> are ineffective. Bisphosphonates, while more efficacious, have an extraordinarily long half-life, which limits their use in adolescents and young women of a reproductive age.<sup>23</sup> Luckily, therefore, oestrogen replacement therapy with transdermal 17b-E2 and cyclic progesterone has been shown to increase spine and hip bone accrual rates in adolescents with AN to roughly equal those of normal-weight controls.<sup>24</sup>

We present here the case of a non-athlete patient with AN and excessive compulsive exercising who developed a pubic stress fracture, which was successfully managed by a combined medical/cognitive-behavioural approach.

### CASE PRESENTATION

A 28-year-old woman with AN was admitted to the Eating Disorder Inpatient Unit of Villa Garda



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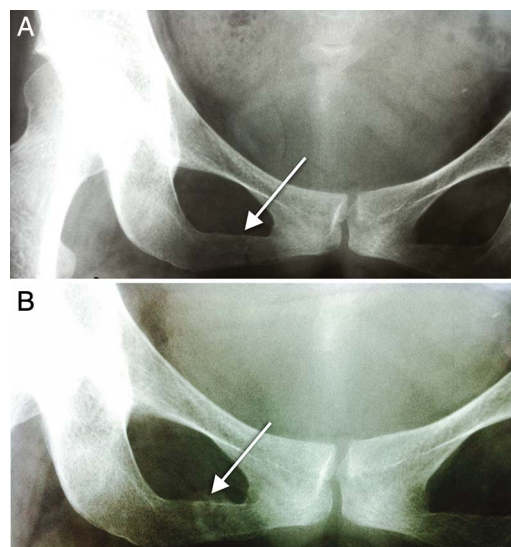
Hospital, Italy, on 2 January 2012. The patient reported that her eating problem had begun at the age of 22 years, when she was abandoned by her boyfriend and lost her job. To cope with the resultant feeling of loneliness, she started to attend a gym daily. At that time she had a body weight of 48 kg and a BMI of 18.9 kg/m<sup>2</sup>. The increased physical activity produced a non-intentional weight loss of about 2 kg in 1 month, and prompted her to pay greater attention to the shape of her body, in particular her legs. Gradually, she became extremely concerned about her weight and shape, and this preoccupation was associated with an increase in the time spent in the gym, and the adoption of a strict low-calorie diet. These extreme weight control behaviours produced a rapid weight loss, and secondary amenorrhoea at a body weight of 44 kg. In roughly 6 months she had reached her lowest weight of 28 kg.

In the 2 years that followed, the patient was repeatedly admitted to medical units in the attempt to stabilise her medical condition. She also attended several specialist outpatient eating disorder treatment programmes, but none of these interventions produced any noticeable improvement in either her body weight or her eating disorder psychopathology. Her body weight eventually normalised after 4 months of hospitalisation in a specialist inpatient eating disorder unit, and she maintained a BMI of about 19 kg/m<sup>2</sup> for three consecutive years after discharge. This she achieved by strictly adhering to the diet prescribed by inpatient team, despite her morbid fear of weight gain and continued excessive compulsive exercising at the gym. However, after this behaviour failed to prevent a slight further increase in body weight, she once again adopted a strict very-low-calorie diet, losing 12 kg in the 6 months before we met her. She was referred to us by her family doctor, and after two assessment sessions agreed to be admitted to our inpatient eating disorder unit.

## INVESTIGATIONS

At admission, the patient had a body weight of 39 kg (BMI 15.2 kg/m<sup>2</sup>), an oral temperature of 35.9°C, a heart rate of 60 bpm, and a blood pressure of 70/40 mm Hg, but no evidence of any orthostatic changes. The following laboratory test results were outside the normal range: alanine aminotransferase 96 IU/L (normal values 14–54 IU/L), alkaline phosphates 138 IU/L (normal values 32–91 IU/L), lactate dehydrogenase 580 IU/L (normal values 266–500 IU/L), ferritin 4.6 ng/mL (normal values 11–306 ng/mL), haemoglobin 10.5 g/L (normal values 12–15.8 g/L), 25OHD 24.2 ng/mL (normal values 30–100 ng/mL), 24 h urinary cortisol 445.5 µg/24 h (normal values 28–213 µg/24 h). Bone density, measured by means of dual-energy X-ray absorptiometry at the femur, was 0.767 g/cm<sup>2</sup>, with evident osteopaenia (t score -1.9). Objective assessment of physical activity using the Sense Wear Pro 2 Armband (SWA; BodyMedia Inc, Pittsburgh, Pennsylvania, USA) during the first week of treatment revealed 206 min/day of physical activity, 11 026 steps/day and a physical activity intensity of 1.8 metabolic equivalent of tasks.

One week after admission, the patient reported a slight pain in the buttock, perineal and thigh regions, and an inability to climb the stairs. Pelvic X-ray showed an irregular, thin fissure at the level of the right ischiopubic ramus, indicating an undisplaced fracture (figure 1A), and excluded the other possible causes of pain in this body district (ie, muscle injuries, lumbar disc disease and gastrointestinal or genitourinary diseases). The consultant orthopaedic specialist recommended rest (lying down) to promote fracture healing.



**Figure 1** Plain radiograph shows (A) non-displaced stress fracture with (B) callus formation at the inferior ischiopubic ramus in a 28-year-old woman with anorexia nervosa.

## TREATMENT

Although the patient had voluntarily accepted admission to our inpatient eating disorder unit, she was very ambivalent towards the treatment, and maintained her excessive compulsive exercising during the first week, as revealed by the SWA assessment.

After receiving the diagnosis of pubic stress fracture, the patient became even more uncooperative, refusing to lie down as recommended by the orthopaedic consultant, or to eat the planned meals, using her inability to walk (as she was able to do before the fracture) to justify her decision. This behaviour was associated with a weight loss of 1 kg in 1 week, and a deterioration in medical conditions and eating disorder psychopathology. At this time the patient also expressed a desire to interrupt the treatment.

The main efforts of the team were therefore focused on motivating the patient not to interrupt the treatment, attempting to engage her in actively addressing her eating disorder psychopathology, and accepting to rest, which would be vital to promote fracture healing. To achieve these goals, we used the following procedures and strategies derived from the 'enhanced' form of cognitive behavioural therapy (CBT-E) for eating disorders used in our unit and described in detail elsewhere.<sup>12 25</sup> Specifically, we first tried to help the patient to choose to accept relative rest and interrupt her excessive compulsive exercising, rather than imposing the decision on her. This was a particularly difficult task because the patient did not see her exercising as a problem, rather as a positive way to control her shape and weight and/or modulate her mood. Nevertheless, by following the key principles of motivational enhancement approaches,<sup>26</sup> in two consecutive sessions we managed to bring her around. This was achieved by first validating her experience by bringing to her attention her perceived positive effect of exercising and ambivalence to change. We informed her of the negative aspects of this form of exercising (ie, maintenance of eating disorder, worsened fracture prognosis, increased risk of other fractures and/or other potential medical complications) and the benefits of rest for fracture healing. Duly informed, the patient was then assisted in drawing up a 'Pros and Cons of Change' table and evaluating her reasons for and against change. During this discussion, we focused the patient on her life aspirations (long-

term goals), not just on the present and positively reinforced every reason for change. We helped the patient to reach the conclusion that the positive aspects of excessive compulsive exercising are of brief duration and invariably associated with negative emotions (eg, guilt if an exercise session is missed), and, in this case, with the high risk of poor recovery from the pelvic stress fracture. To address the patient's fear of losing control over her weight if she interrupted exercising, we reassured her that the adoption of a healthy lifestyle is the best way to maintain long-term weight control. Finally, we guided the patient to the conclusion that changing her unhealthy exercising practice would be a positive opportunity for a new start, a life no longer conditioned by the need to exercise. Since the patient remained reluctant, we suggest that she 'take the plunge' and make a fresh start on a trial basis. We informed her that over the first few days, her anxiety levels and concern about shape and weight would be likely to increase, but that these would gradually diminish as the treatment progressed, along with an associated gain in the benefits of leading a healthy lifestyle. The procedures above helped her to make the decision to continue, and to address her urge to exercise, although we cannot exclude the influence of at least two other factors in facilitating the patient's engagement in the treatment. First and foremost, the objective ischiopubic fracture may have made the patient more aware of how harmful AN can be. Second, the patient was managed by highly trained clinical staff via specific strategies and steps in an environment designed to promote trust and collaboration.

Once the patient was back on track, the next step in the programme was to ask her to keep a real-time monitoring record of the frequency, duration and type of exercise she engaged in. We emphasised that writing this down in real-time is a key factor for change,<sup>25</sup> because becoming aware of what she was doing, thinking and feeling at the precise time that she was exercising would enable her to see that she could choose to change her behaviour, even if this at first seemed automatic and outside her control.

Our third major intervention was to suggest that the patient address her urge to exercise by means of the 'Things to Say and Do' procedure. This comprises drawing up a list of tasks that the patient can refer to when she feels the temptation to exercise. For example, 'Things to Do' includes activities that are incompatible with exercising (eg, taking a bath, doing a crossword, knitting) or reduce the risk thereof (eg, staying in the living room with other patients), while 'Things to Say' comprises external vocalisations of statements reiterating, for instance, that the urge to exercise is a temporary phenomenon that can be tolerated and overcome.

### OUTCOME AND FOLLOW-UP

The patient gradually became actively engaged in the treatment. She rested consistently as recommended, and X-ray (figure 1B) confirmed complete fracture healing in 35 days. Moreover, she addressed her low body weight and undereating through assisted meals, and the other features of her eating disorder psychopathology were dealt with in the scheduled individual and group CBT-E sessions. In the last part of the treatment, administered in day hospital, she also began healthy social exercising (ie, tennis lessons). After a total of 20 weeks (13 inpatient and 7-day-hospital) the patient was discharged on 20 May 2012 with a body weight of 47.5 kg and a BMI of 18.8 kg/m<sup>2</sup>. Following discharge, the patient completed a 6-month course of outpatient CBT-E treatment (until November 2012), and at 12-month follow-up (in May 2013) she had a body weight of 51 kg, a BMI of 20.1 kg/m<sup>2</sup>, regular menses, complete remission of the eating disorder psychopathology, good social relationships and a job as an au pair. Despite these promising signs, the late onset

of the eating disorder and the patient's history of relapse after a period of maintaining normal body weight without improvement in her eating disorder psychopathology, mean that definitive conclusions about her outcome may only be drawn after longer-term monitoring.

### DISCUSSION

Stress fractures are a difficult to manage complication of AN when it coexists with excessive compulsive exercising, but to date little indication has been provided in the literature on how best to manage such patients. Hence we describe the main CBT-E-based procedures and strategies we used successfully in a non-athlete AN patient with excessive compulsive exercising and pubis fracture (other specific procedures and strategies that can be used to address excessive compulsive exercising in eating disorders have been reported by the Loughborough Eating Disorders Activity Programme, LEAP).<sup>27</sup>

Stress fractures in AN individuals commonly occur at the femoral neck,<sup>28 29</sup> the heel,<sup>30</sup> and the proximal fibula,<sup>31</sup> but to our knowledge, stress fracture of the pubis in AN has only previously been described in one young adult male athlete.<sup>32</sup> Nevertheless, this case report should alert clinicians to the need to pay special attention to patients with AN, even if they are not osteoporotic, who present other risk factors for stress fractures such as excessive compulsive exercising, low serum levels of 25OHD, iron deficiency and amenorrhoea. When patients with AN and excessive compulsive exercising present with lower back, lumbar, buttock, inguinal or thigh pain, however slight, clinicians should suspect the presence of pelvic stress fracture. In such patients, specific procedures and strategies derived from CBT-E for eating disorders used to treat their eating disorder psychopathology can be tailored to help encourage rest, without coercive measures, in order to promote healing.

### Learning points

- ▶ Inferior ischiopubic stress fracture is a complication that may occur, even during hospitalisation, in patients with anorexia nervosa who practice excessive and compulsive exercising.
- ▶ Osteoporosis is not the only risk factor for this fracture, and clinicians must assess for others, such as excessive and compulsive exercising, low serum levels of 25-hydroxyvitamin D, iron deficiency and amenorrhoea.
- ▶ Slight insidious pain over the groin, perineal region, buttock or thigh must raise suspicions of pelvic fracture.
- ▶ Pelvic X-ray is usually sufficient for diagnosis, although pelvic MRI may be necessary in some cases.
- ▶ Specific cognitive behavioural procedures, rather than coercive measures, can be used to encourage patients with anorexia nervosa, excessive compulsive exercising and stress fracture to rest, thereby promoting healing while addressing the underlying eating disorder psychopathology.

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