STRESS FRACTURE OF THE INFERIOR AND SUPERIOR PUBIC RAMUS IN A MAN WITH ANOREXIA NERVOSA AND HYPOGONADISM

E. THIENPONT, J. BELLEMANS, I. SAMSON, G. FABRY

The authors present the case of a 23-year-old athlete with a bifocal stress fracture of the pubis. Bifocal cases are rather rare, therefore an insufficiency fracture was suspected. Further investigation showed osteopenia due to secondary hypogonadotropic hypogonadism caused by anorexia nervosa. This case illustrates the need for awareness of underlying metabolic disorders in very rare cases of stress fractures in athletes.

Keywords: stress fracture; pubic ramus; hypogonadism; anorexia nervosa.
Mots-clés: fracture de fatigue; branche pubienne; hypogonadisme; anorexie nerveuse.

Stress fractures occur as a result of repeated cyclic loading of bone (6). Depending on the condition of that bone, they can be divided into two types:

(1) fatigue fractures which occur in bone with normal elastic resistance that is loaded to an unaccustomed degree

and

(2) insufficiency fractures which occur in weakened bone (6). Osteoporosis, rheumatoid arthritis, previous irradiation and hormonal or metabolic disorders may be predisposing factors (2, 3).

The most commonly affected bones are the metatarsals and calcaneus, followed by the tibia and fibula. Stress fractures of the pubic ramus, especially bifocal fractures, are seen less often (3, 5).

CASE REPORT

A 23-year-old long-distance runner and physiotherapist presented at the outpatient clinic with pain in the left groin and ischial tuberosity. The problem started three weeks earlier while he was running. He took some pain medication and continued to run. He consulted our department because the pain didn’t diminish.

His history revealed an excessive drive for physical activity (long-distance running and bicycling) caused by anorexia nervosa. There was no self-induced vomiting or use of laxatives and diuretics. Neither was there a family history of osteoporosis, recurrent fractures or metabolic disorders.

Physical examination revealed a man with a height of 175 cm and a weight of 53 kg (Body Mass Index = 16, normal value = 20-25). His blood pressure was 90/60 mm Hg, and the pulse rate was 48 beats/min. There was discomfort in the groin when standing unsupported on one foot (‘positive standing sign’), and deep palpation revealed extreme tenderness localized to the pubic ramus and ischial tuberosity.

X-ray of the pelvis showed unfused physes and a fracture of the left inferior pubic ramus with periosteal callus formation. A bone scan demonstrated areas of increased radionuclide uptake in the left inferior and superior pubic ramus and also in the left femoral neck.

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Because of the unusual bifoal lesion a CT-scan and bone densitometry were performed. The CT-scan confirmed the fracture without providing arguments for tumor activity. The lesion in the femoral neck was most likely fibrous dysplasia.

Bone density measured by computed tomography at the lumbar spine (0.832 g/cm² as the mean value for L₁ through L₄ vs. 1.140 g/cm² for an age/sex-matched control) and by single-photon absorptiometry at the distal radius (0.656 g/cm² as the mean value vs. 0.800 g/cm² for an age/sex-matched control) both showed osteopenia. Because of this osteopenia and anorexia nervosa, the patient was referred to the department of endocrinology. History revealed here that he only shaved every three days and had occasional erections but no ejaculations. Physical examination showed Tanner Stage II pubic hair. There were normal penis size but small and soft testes (4 ml), thyroid size was normal.

Biochemical analysis showed slight leukopenia (2.4 × 10⁹/l) and anemia (4 × 10⁹/l). The levels of serum testosterone (77 mg/dl) and free testosterone (0.98 ng/dl) were low, and luteinizing hormone (1.8 mU/ml) and follicle-stimulating hormone (< 10 mU/ml) were inappropriately normal. Responses of LH and FSH to a bolus of gonadotropin-releasing hormone were blunted. The 24-hour urine level of free cortisol (162 μg/24h) and the dosage for growth hormone (9.7 μIU/ml) were both elevated.

For somatomedin C (108.3 ng/ml), ACTH (28 pg/ml) and free cortisol (1.19 μg/dl) normal values were found (table I).

The serum cortisol level was adequately suppressed following administration of 1 mg dexamethasone. MRI of the sella turcica showed no pathologic processes.

Therapy was started with choriongonadotropin (HCG) 3 × 1500 IE intramuscular injections and psychiatric counseling. This patient gained 3 kg in 2 months and evolved to Tanner stage IV. His testes became bigger (10 ml left and 14 ml right), and the testosterone level increased to a high value (924 ng/dl).

The pubic stress fractures both healed completely after he stopped all physical activity.
Fig. 3. — On the left side CT-scan shows the pubic fracture with surrounding callus formation.
On the right side CT-scan confirms fibrous dysplasia of the femoral neck.

Table 1. — Biochemical evaluation at time of diagnosis

<table>
<thead>
<tr>
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<th>Case</th>
<th>Normal range</th>
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<tbody>
<tr>
<td>Testosterone ng/dl</td>
<td>77</td>
<td>300-1000</td>
</tr>
<tr>
<td>Free testosterone ng/dl</td>
<td>0.98</td>
<td>5-20</td>
</tr>
<tr>
<td>Basal FSH mIU/ml</td>
<td>&lt;1.0</td>
<td>2-10</td>
</tr>
<tr>
<td>Basal LH mIU/ml</td>
<td>1.8</td>
<td>2-10</td>
</tr>
<tr>
<td>Estradiol pg/dl</td>
<td>10</td>
<td>0-30</td>
</tr>
<tr>
<td>Progesterone µg/dl</td>
<td>0.09</td>
<td>0-0.2</td>
</tr>
<tr>
<td>Thyroxine µg/dl</td>
<td>8.3</td>
<td>5.5-12.0</td>
</tr>
<tr>
<td>Triiodothyronine resin uptake %</td>
<td>98.8</td>
<td>90-118</td>
</tr>
<tr>
<td>Total triiodothyronine ng/dl</td>
<td>85.5</td>
<td>80-190</td>
</tr>
<tr>
<td>Thyrotrpin mIU/l</td>
<td>1.8</td>
<td>0.5-3.5</td>
</tr>
<tr>
<td>Albumin g/dl</td>
<td>4.93</td>
<td>3.35-4.57</td>
</tr>
<tr>
<td>Alkaline phosphatase U/l</td>
<td>173</td>
<td>90-260</td>
</tr>
<tr>
<td>Calcium mg/dl</td>
<td>10.10</td>
<td>8.9-10.5</td>
</tr>
<tr>
<td>Inorganic phosphorous mg/dl</td>
<td>4.3</td>
<td>2.3-4.7</td>
</tr>
<tr>
<td>Calciumin pg/ml</td>
<td>27</td>
<td>0-28</td>
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<tr>
<td>Osteocalcine ng/ml</td>
<td>6.4</td>
<td>11-43</td>
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<tr>
<td>Parathyroid hormone pg/ml</td>
<td>18.0</td>
<td>3-40</td>
</tr>
<tr>
<td>25-Hydroxyvitamin D ng/ml</td>
<td>47.3</td>
<td>7-60</td>
</tr>
<tr>
<td>Serum cortisol µg/dl</td>
<td>19.4</td>
<td>10-22</td>
</tr>
<tr>
<td>Free cortisol µg/dl</td>
<td>1.19</td>
<td>0.7-1.8</td>
</tr>
<tr>
<td>Growth hormone µU/ml</td>
<td>9.7</td>
<td>0-7.5</td>
</tr>
<tr>
<td>Somatomedin C ng/ml</td>
<td>108.3</td>
<td>100-300</td>
</tr>
<tr>
<td>ACTH pg/ml</td>
<td>28</td>
<td>0-80</td>
</tr>
<tr>
<td>Prolactin µU/ml</td>
<td>&lt;110</td>
<td>0-1000</td>
</tr>
<tr>
<td>White Blood Cells 10^9/l</td>
<td>2.4</td>
<td>4.0-11.0</td>
</tr>
<tr>
<td>Red Blood Cells 10^9/l</td>
<td>4</td>
<td>4.5-5.9</td>
</tr>
<tr>
<td>Ferritin µg/l</td>
<td>14</td>
<td>16-330</td>
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<tr>
<td>Vit B12 ng/l</td>
<td>532</td>
<td>170-800</td>
</tr>
<tr>
<td>Folate µg/l</td>
<td>5.5</td>
<td>2.5-110.0</td>
</tr>
<tr>
<td>Urine free cortisol µg/24h</td>
<td>162</td>
<td>&lt; 100</td>
</tr>
<tr>
<td>Urinary calcium mg/24h</td>
<td>186</td>
<td>&lt; 300</td>
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</table>

**DISCUSSION**

Pubic stress fractures are uncommon problems in athletes (only 6%) (3, 4). However they are most often seen in long-distance runners and jumpers (5). This is because the pubis is exposed to tensile stresses in alternating fashion during the stance and swing phases. During the first hip adductors steady the femur against the pull of the hip abductors, while during the latter the external rotators contract to compensate for the rotation of the pelvis toward the opposite limb. Both work on either side of the ischiopubic line (1, 4, 5).

Predisposing factors for stress fractures are Caucasian race, lack of physical conditioning, mixed training of men and women, osteopenia, metabolic and biochemical disorders, youth and female gender (1, 3, 4, 5).

Indeed pubic stress fractures occur more commonly in young athletes and in women (1, 4, 5). In females the bones are more slender, the margins of the ischiopubic rami are less everted and the obturator foramen is more triangular than oval (1, 3, 4, 5). Also there could be more hip-extension forces during their running pattern, generating more tensile stresses in the medial portion of the pubic ramus (3, 4, 5).

In this case history and physical examination were quite typical (3). To differentiate between adductor tendinitis, muscle tears in adductors and
hamstrings, osteitis pubis and avulsion fractures of
the adductor or hamstring muscles, an xray and
radionuclide bone scan (Tc 99MDP) are neces-
sary (3, 4).

Pubic stress fractures can present on xray in an
occult or in an agressive way (2, 6). For occult frac-
tures further investigation with a bone scan is
mandatory (3, 4, 5, 6). A technetium bone scan is
also very useful to evaluate the sacroiliac joint (3,
6). Due to the anatomy of the pelvic ring, if one of
the three joints is unstable, shearing forces are cre-
ated and stress is transmitted to other portions of
the ring.

Therefore instability at the sacroiliac joint
caused by chronic stress can lead to secondary
stress reactions at the symphysis pubis (3). Pubic
instability too can lead to sacral stress fractures (3).
In these cases the history is often that of sciatica,
and plain films can show sclerosis, erosions or
osteophytes at the sacroiliac joint, but very often
they show nothing (3, 6). In these cases bone scan
can focus our attention on the posterior joint, and
CT-scan or MRI can confirm the sacral stress frac-
ture (3, 6).

If a pubic stress fracture presents in an agressive
way, malignancy has to be excluded (2, 6). For this
we prefer CT-scan since it shows the fracture line
and surrounding callus formation (6). The use of
MRI can lead to incorrect conclusions since the
image is difficult to differentiate from low-grade
chondrosarcoma (2). On T2-images we see a very
hyperintense mass lesion with a hypointense rim
showing peripheral and sepal enhancement mim-
icking the ‘ring and arc’ from low grade chon-
drosarcoma after contrast administration (2).

The pathologist too can support this wrong diag-
nosis, since a biopsy would show hemorrhage,
fibrosis, woven bone, dead bone and cartilage (2).
This could suggest osteomyelitis, osteosarcoma or
chondrosarcoma (2). The MRI findings could thus
be sustained by the pathology and lead to overtreat-
ment and dramatic consequences.

Bone densitometry is very useful to objectivate
insufficiency fractures. Often osteopenia is difficult
to evaluate on plain xrays. Although athletes are in
very good physical condition, osteopenia is often
seen. The cases of female athletes with hypothala-
mic amenorrhea and estrogen deficiency which
leads to bone demineralisation are well known (4).
The effect of exercise on male gonadal function has
been less well studied. Reduced testosterone levels
have been reported in longdistance runners, which
may result from alterations in the hypothalamic-
pituitarygonadal axis. However, although detailed
studies are lacking, the observed reductions in
testosterone levels are not as marked as those
occurring in male anorectics (7).

The relationship between skeletal mass in male
runners and their gonadal function has not been
investigated. However it is well documented that
male endurance athletes have greater bone mass
than sedentary men and that amenorrheic women
athletes have a lower bone mass than sedentary or
athletic women with menses, but greater than
sedentary hypogonadal women (7).

Running thus has an ambiguous effect. While it
is stimulating the bone formation on one side, it is
possibly inducing several changes at the hormonal
level and definitely is producing a further weight
loss. Indeed in this case our patient is not only a
long-distance runner but is also suffering from
anorexia nervosa.

Anorexia nervosa is a disorder that primary
affects young women and is characterised by self-
induced weight loss, psychological disturbances
and a complex array of secondary physiologic
alterations. Only 5% to 10% of cases are seen in
males. Osteopenia in anorectics is a well known
complication (7). It results first of all from the
hypogonadotropic hypogonadism. Although much
less is known about the role of gonadal steroids and
osteopenia in males, states of prolonged testoster-
one deficiency, such as occur in Kallmann’s syn-
drome and Klinefelter’s syndrome, are associated
with osteopenia (7). Secondly we know that
somatomedin C levels are reduced in circum-
stances of fasting and protein-calorie malnutrition.
Somatomedin has an anabolic effect on bone cells
and chondrocytes and also stimulates the incorpo-
ration of sulfate into cartilage. Animal models of
starvation have shown a fall in growth cartilage in
association with reduced somatomedin levels, both
of which are reversible with refeeding (7). Finally
anorectics have increased cortisol production rates,
elevated serum cortisol levels and abnormal results of dexamethasone suppression tests (7). Cortisol excess is known to be associated with bone demineralisation (7). Both cortisol and somatomedin levels were normal in this case, and low-dose dexamethasone suppression was also normal, but free urinary cortisol remained elevated, indicating absence of normal rhythm and exposure of the tissue to excessive cortisol levels.

In anorectics no apparent disorders in the calcium or vitamin D metabolism were found (7), this was also the case in our patient.

Therefore treatment of the anorexia and secondary hypogonadism was mandatory, since other stress fractures would have presented after periods of rest. Normally this is sufficient for union of the fracture to occur (1, 3, 5). Delayed union or non-union are rarely seen (4). If no progression is seen after long periods of rest, surgery with bone grafting may be necessary (3, 4, 5).

CONCLUSION

This case proves that even when the x-rays clearly show a suspected stress fracture, additional screening with a bone scan is useful. Furthermore the need for a biochemical and endocrinological analysis in athletes is demonstrated. The need for awareness of insufficiency fractures in the young and healthy athlete is thereby shown.

REFERENCES


SAMENVATTING

E. THIENPONT, J. BELLEMANS, I. SAMSON, G. FABRY. Stressfractuur van de onderste en bovenste pubistak bij een man met anorexia nervosa en hypogonadisme.

De auteurs beschrijven een bifocale stressfractuur van de pubis bij een 23-jarige atleet. Bifocale stressfracturen zijn eerder zeldzaam en daarom werd een insufficiëntief fractuur vermoed. Verdere investigatie toonde osteopenie aan, secundair aan hypogonadotroop hypogonadisme tengevolge van anorexia nervosa. Dit geval illustreert dat men bedacht dient te zijn op onderliggend metabool lijden in zeldzame gevallen van stressfracturen bij atleten.

RÉSUMÉ

E. THIENPONT, J. BELLEMANS, I. SAMSON, G. FABRY. Fracture de fatigue des branches supérieure et inférieure du pubis chez un homme atteint d'anorexie nerveuse et d'hypogonadisme.

Les auteurs présentent un cas de fracture de fatigue bifocale du pubis chez un athlète de 23 ans. Les fractures de fatigue bifocales sont plutôt rares et pour cette raison, on a suspecté une fracture par insuffisance. Une investigation plus poussée a montré une ostéopénie secondaire à un hypogonadisme causé par une anorexie nerveuse. Ce cas nous apprend qu’il faut rechercher des problèmes métaboliques dans le cas de fractures de fatigue inhabituelles chez des athlètes.