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.

Xray 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.

Department of Orthopedic Surgery, University Hospital Pellenberg, K.U. Leuven, Belgium.
Correspondence and reprints : J. Bellemans, U.Z. Pellenberg, Weligerveld 1, B-3212 Pellenberg, Belgium.

Fig. 1. — Plain radiography reveals callus formation at the left inferior pubic ramus and unfused physes at the femoral head.

Fig. 2. — Radionuclide bone scan shows increased uptake at the left inferior and superior pubic ramus, and slightly increased uptake at the femoral neck region.

Because of the unusual bifocal 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/cm^2 as the mean value for L_2 through L_4 vs. 1.140 g/cm^2 for an age/sex — matched control) and by single-photon absorptiometry at the distal radius (0.656 g/cm^2 as the mean value vs. O.800 g/cm^2 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 (2.4×10^9/l) and anemia (4×10^{12}/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 chorionogonadotropin (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.

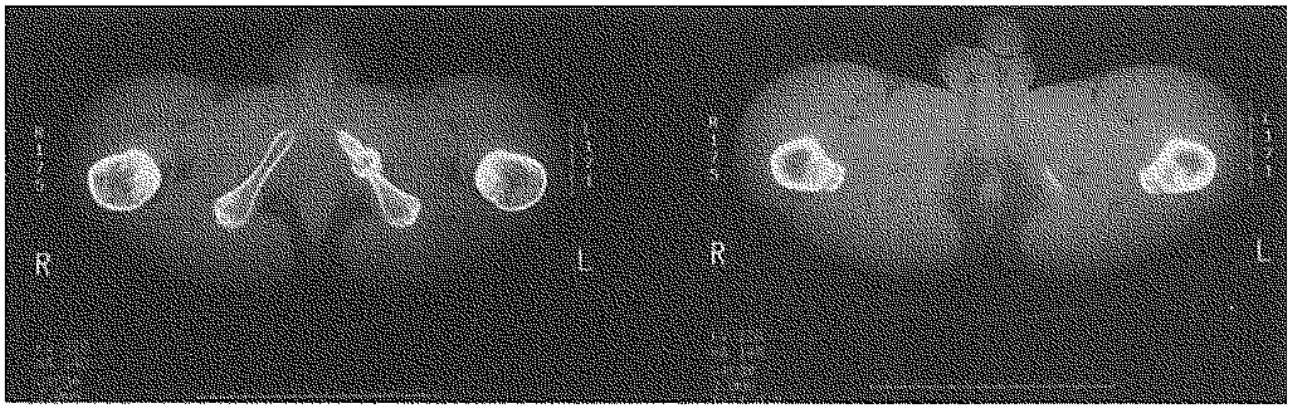

Fig. 3. — On the left side CT-scan shows the pubic fracture with surrounding callus formation. On the right side CT-scan confirms fibrous dysplasia of the femoral neck.

Table I. — Biochemical evaluation at time of diagnosis

	Case	Normal range
Testosterone ng/dl	77	300-1000
Free testosterone ng/dl	0.98	5-20
Basal FSH mU/ml	< 1.0	2-10
Basal LH mU/ml	1.8	2-10
Estradiol pg/ml	10	0-30
Progesterone µg/dl	0.09	0-0.2
Thyroxine µg/dl	8.3	5.5-12.0
Triiodothyronine resin uptake %	98.8	90-118
Total triiodothyronine ng/dl	85.5	80-190
Thyrotropin mU/l	1.8	0.5-3.5
Albumin g/dl	4.93	3.85-4.57
Alkaline phosphatase U/l	173	90-260
Calcium mg/dl	10.10	8.9-10.5
Inorganic phosphorus mg/dl	4.3	2.3-4.7
Calcitonin pg/ml	27	0-28
Osteocalcine ng/ml	6.4	11-43
Parathyroid hormone pg/ml	18.0	3-40
25-Hydroxyvitamin D ng/ml	47.3	7-60
Serum cortisol µg/dl	19.4	10-22
Free cortisol µg/dl	1.19	0.7-1.8
Growth hormone µIU/ml	9.7	0-7.5
Somatomedin C ng/ml	108.3	100-300
ACTH pg/ml	28	0-80
Prolactin µU/ml	< 110	0-1000
White Blood Cells 10^9/l	2.4	4.0-11.0
Red Blood Cells 10^{12}/l	4	4.5-5.9
Ferritin µg/l	14	16-330
Vit B12 ng/l	532	170-800
Folate µg/l	5.5	2.5-11.0
Urine free cortisol µg/24h	162	< 100
Urinary calcium mg/24h	186	< 300

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 the 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 necessary (3, 4).

Pubic stress fractures can present on xray in an occult or in an agressive way (2, 6). For occult fractures 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 created 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 fracture (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 septal enhancement mimicking the 'ring and arc' from low grade chondrosarcoma after contrast administration (2).

The pathologist too can support this wrong diagnosis, 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 overtreatment 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 hypothalamic 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 testosterone deficiency, such as occur in Kallmann's syndrome and Klinefelter's syndrome, are associated with osteopenia (7). Secondly we know that somatomedin C levels are reduced in circumstances of fasting and protein-calorie malnutrition. Somatomedin has an anabolic effect on bone cells and chondrocytes and also stimulates the incorporation 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 nonunion 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 xrays 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

1. Hill P., Chatterji S., Chambers D., Keeling J. Stress fracture of the pubic ramus in female recruits. J. Bone Joint Surg., 1996, 78-B, 383-386.
2. Hosono M., Kobayashi H., Fujimoto R., Tsutsui K., Kotoura Y., Tsuboyama T., Hayashi H., Nakamura T., Konishi J. MR appearance of parasymphyseal insufficiency fractures of the os pubis. Skeletal Radiol., 1997, 26, 525-528.
3. Major N., Helms C. Pelvic stress injuries : The relationship between osteitis pubis (symphysis pubis stress injury) and sacroiliac abnormalities in athletes. Skeletal Radiol., 1997, 26, 711-713.
4. O'Brien T., Wilcox N., Kersch T. Refractory pelvic stress fracture in a female long-distance runner. Am. J. Orthop., 1995, 24, 710-713.
5. Pavlov H., Nelson T., Warren R., Torg I., Burstein A. Stress fractures of the pubic ramus : A report of twelve cases. J. Bone Joint Surg., 1982, 64-A, 1020-1025.
6. Peris P., Guanabens N., Pons F., Herranz R., Monegal A., Suris X., Munoz-Gomez J. Clinical evolution of sacral stress fractures : Influence of additional pelvic fractures. Ann. Rheum. Dis., 1993, 52, 545-547.
7. Rigotti N., Neer R., Jameson L. Osteopenia and bone fractures in a man with anorexia nervosa and hypogonadism. JAMA, 1986, 256, 385-388.

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 insufficiëntiefractuur 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 plûtot 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.