

Original Article

Fatigue sacral fractures: A case series and literature review

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Abstract

Objectives: Fatigue sacral fractures (FSFs) are rare and often misdiagnosed. This study presents a series of FSFs and a meticulous literature review. **Methods:** The present is an 11-year (2010-2021) retrospective observational study. The characteristics of all adult patients with FSF, including demographics, fracture type, treatment, history of fatigue fracture and imaging were evaluated. **Results:** Eight cases (6 females; 75%), suffering from 12 fractures (4 bilateral cases) with mean age=33.4 years were studied. Two patients (25%) had suffered another fatigue fracture in the past. Mean symptoms' duration prior diagnosis was 8.5 weeks, while mean symptoms' duration after diagnosis was 10.75. In most cases (7; 87.5%), MRI revealed the fracture. According to the Kaeding-Miller classification; five fractures (42%) were grade III, four (33%) IV and three (25%) II. All patients were treated conservatively, with rest and analgesics, while three received vitamin D and calcium. One patient, due to delayed union, was commenced on teriparatide. **Conclusions:** FSFs are often misdiagnosed; therefore, they should be included in the differential diagnosis for chronic low back-or-hip pain in athletes. History of other fatigue injuries seems to be a predisposing factor. It is of paramount importance to obtain advanced imaging for identifying a FSF.

Keywords: Fatigue Fracture, Fatigue Fractures in Athletes, Sacral Fracture, Teriparatide, Vitamin D

Introduction

Stress sacral fractures are most commonly presented with low-back or pelvic pain and are divided into two main categories: insufficiency and fatigue fractures¹. Those are distinguished according to mechanism of injury and osseous physiology. Insufficiency fractures occur to decreased density bone with the application of normal forces, while fatigue fractures occur to normal bone with the application

of repeated stresses¹. Insufficiency fractures are most commonly observed in elderly patients, while fatigue fractures in young athletes^{1,2}.

Fatigue sacral fractures (FSFs) are rare, since only a few reports have been described so far, and often misdiagnosed due to clinical entities with similar symptomatology, such as sacroiliitis, vertebral fractures, degenerative disk disease, trochanteric bursitis, spondylolisthesis, facet arthropathy and low back muscle strains¹⁻³. These injuries may occur in both males and females athletes, as well as military recruits, while they have been associated with a plethora of sports activities, such as long-distance running, soccer, weightlifting and badminton^{2,3}.

This study presents a case series of FSFs, as well as a meticulous literature review regarding imaging and management, aiming to increase the awareness of physicians for these injuries in cases of low-back and/or pelvic pain in young active patients.

The authors have no conflict of interest.

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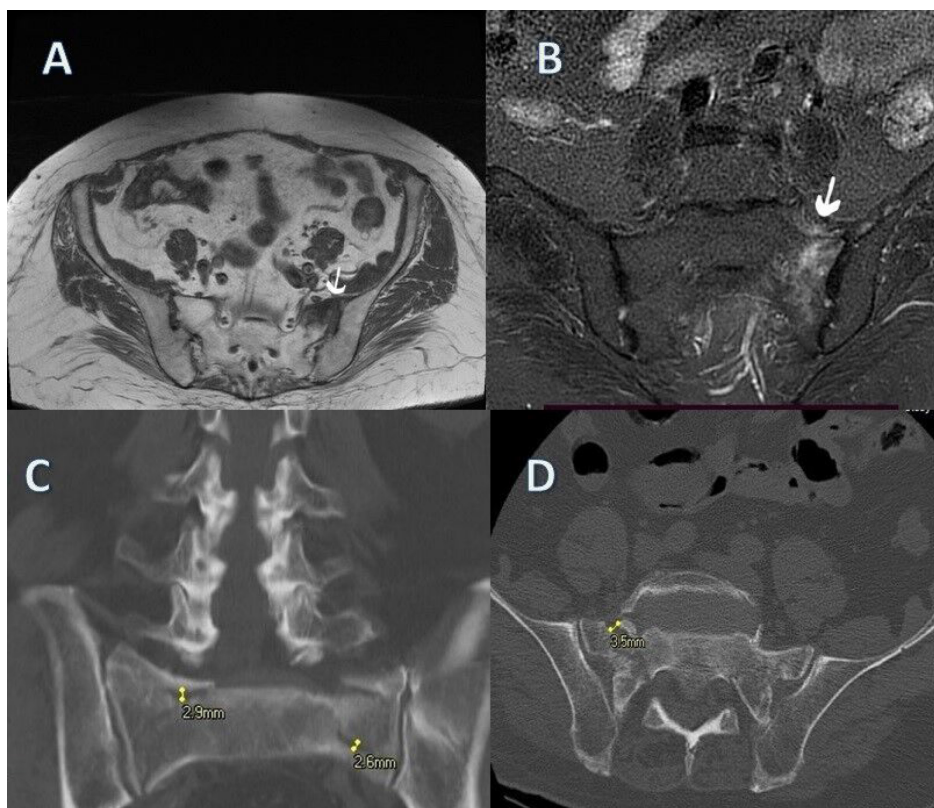


Figure 1. A. T1 weighted MRI axial views of a fatigue sacral fracture (case 1 in Table 1). B. STIR MRI coronal view of the same fracture (case 1 in table 1). Fracture is classified according to Kaeding Miller classification as grade III. C coronal CT view of bilateral sacral fractures of case 2 (Table 1), D axial CT view of the same fractures. Both fractures are classified as grade IV according to Kaeding Miller classification.

Materials and Methods

The present is a retrospective observational study of a prospectively maintained database. All patients treated with a FSF at the Department of Orthopaedics and Traumatology of the “Venizeleion” General Hospital of Heraklion, Crete, Greece from January 2010 until January 2021 were eligible to participate. Exclusion criteria patients suffering from insufficiency sacral fractures.

The “Venizeleion” General Hospital is a 600-bed secondary hospital on the island of Crete, Greece. It represents one of the two big reference hospitals of the whole island, serving a population of approximately 650,000, who is mostly rural and to a lesser extend urban and suburban.

Patients’ demographics, fracture site and type, type of sports activity, duration of symptomatology prior to diagnosis, imaging, management, duration of symptomatology after diagnosis, history of fatigue fractures and follow-up were evaluated from the medical records. The fracture site was divided into the sacral corpus and alar.

Fractures were classified according to the Kaeding-Miller classification system. According to this classification, Grade

I indicates asymptomatic stress reaction on imaging, Grade II indicates pain with no fracture line, Grade III indicates non-displaced fracture, Grade IV indicates displaced fracture (>2 mm), while Grade V is consistent with nonunion.

The present study has been approved from the bioethics committee of “Venizeleion” General Hospital of Heraklion, Crete, Greece.

Results

During this 11-year period, a total of 8 cases (6 females; 75% and 2 males; 25%) were included in the study (Table 1). The studied population’s mean age was found to be 33.4 years [standard deviation (SD)=7.5]. Regarding the medical history 2 patients (25 %) had suffered from fatigue fractures in the past; one from tibia and one from metatarsal fracture. Patients’ mean body mass index (BMI) was 21.7 kg/m².

Regarding the type of sports activity, 5 patients were long-distance runners, one played tennis, one soccer and one jogged. The mean duration of symptoms prior to diagnosis was found to be 8.5 weeks (SD=4.5), while the

Table 1. Characteristics of the studied population. Patients' demographics, well as fractures' characteristics are presented. Details about treatment, imaging confirming the diagnosis, history of fatigue fracture duration of symptoms and follow-up are also presented.

Case	Fracture region	Kaeding Miller Classification	Imaging confirming diagnosis	Gender	Age	Type of activity	Duration of symptoms before Diagnosis (weeks)	Treatment	Duration of symptoms after diagnosis (weeks)	History of fatigue Fractures	New fatigue fracture during follow-up	Follow-up (months)
1	Ala	III	MRI	Female	46	Long-Distance Running	8	Rest, Analgesics, Vit D, Calcium	6	-	-	14
2	Corpus	IV (right), IV (left)- (Bilateral)	CT	Male	40	Jogging	5	Rest- analgesics, Vit D, Calcium	12	-	-	38
3	Ala	II (right), II (left)- (Bilateral)	MRI	Female	32	Tennis	4	Rest, analgesics, Partial weight bearing (Crutches) for 3 weeks	6	-	Metatarsal (3 years after diagnosis of sacral fracture)	62
4	Corpus	IV (right), IV (left)- (Bilateral)	MRI	Female	28	Long-Distance Running	12	Rest, analgesics, Vit D, Calcium, Teriparatide (after 16 weeks due to delayed union)	36	Tibia (6 years ago)	-	30
5	Ala	II	MRI	Male	24	Soccer	8	Rest, Analgesics, Physiotherapy	4	-	-	18
6	Ala	III (right), III (left)- (Bilateral)	MRI	Female	33	Lon-Distance Running	7	Rest, Analgesics, Physiotherapy	5	-	-	74
7	Ala	III	MRI	Female	38	Long-Distance Running	18	Partial weight bearing (Crutches), Rest-analgesics	8	-	Tibia (2 years after diagnosis of sacral fracture)	100
8	Ala	III	MRI	Female	26	Long-Distance Running	6	Partial weight bearing (Crutches), Rest-analgesics	9	Metatarsal (3 years ago)	-	42

mean symptoms duration after diagnosis was 10.75 weeks (SD=10.5). The main symptom was low back pain in most cases (6;75%) and hip pain in the remaining 2 (25%).

A total of 12 fractures were found, since 4 patients suffered bilateral sacral fractures. In the majority of the cases (7; 87.5%), diagnosis was established with magnetic resonance imaging (MRI), while in one case (case 2 in Table 1) computer tomography (CT) offered adequate information for diagnosing the fractures (Figure 1).

Eight fractures were located at the sacral ala, while the remaining four at the sacral corpus. According to the Kaeding Miller classification; five fractures (42%) were characterized as grade III, four (33%) as grade IV and three (25%) as grade II.

Treatment included rest and analgesics in all cases, while vitamin D and calcium were given in 3 (37.5%) patients. Partial weight bearing was implemented in another 3 (37.5%), while 2 (25%) were supported with physiotherapy.

In one case (case 4 in Table 1) due to delayed union (12 weeks with persistent pain), the patient was commenced on teriparatide for 24 weeks, while after that period the patient was asymptomatic.

During follow-up, two patients suffered an additional fatigue fracture; one from a metatarsal and the other from a tibia fracture.

Discussion

In 1989 Volpin et al described the first FSFs in healthy military recruits⁴. Even nowadays these injuries are considered relative rare¹⁻³. Fatigue fractures occur to normal bone with the application of repeated forces and have been mainly associated with long distance runners, as well as soldiers with regular weight-lifting activities¹⁻⁴.

Through a meticulous electronic literature review, including the PubMed, Medline and Embase databases, from

Table 2. Characteristics of the reviewed cases (from 2000-2021). Details about patients' demographics, fracture region, imaging confirming the diagnosis, history of fatigue fracture, treatment and duration of symptoms are presented.

Author	Year	Fracture side	Fracture-region	Imaging confirming diagnosis	Gender	Age	Type of activity	Duration of symptoms before Diagnosis (Weeks)	Treatment	Duration of symptoms after diagnosis (Weeks)	History of stress Fractures
1. Klossner ⁶	2000	Right	Ala	Bone scan, CT	Female	19	Long distance running	5	Rest	4	None
2. Lam et al ⁷	2001	Right	Wings	Bone scan, MRI	Male	10	School physical education course	2	Bed rest-analgesics	1	None
3. Johnson et al ⁸	2001	Left	Ala and sacroiliac joint	Bone scan, MRI	Female	21	Soccer	2	Non-weight bearing, calcium, estrogen supplementation	48	Metatarsal, tibia (bilateral)-12 months ago
4. Johnson et al ⁸	2001	Left	Ala	Bone scan, MRI, CT	Female	20	Basketball	5.5	Rest	36	None
5. Johnson et al ⁸	2001	Left	Ala	CT, MRI	Female	45	Running	6	Rest, Calcium, calcitonin, Vit D	56	None
6. Johnson et al ⁸	2001	Left	Ala and sacroiliac joint	Bone scan, MRI, CT	Female	22	Cross country running	0.5	Non-weight bearing	36	None
7. Johnson et al ⁸	2001	Left	Inferior sacroiliac joint	Bone scan, MRI	Female	41	Jogging	20	Rest	20	None
8. Johnson et al ⁸	2001	Right	Superior sacrum	Xrays, Bone scan, MRI	Female	19	Jogging	26	Rest	52	None
9. Johnson et al ⁸	2001	Right	Ala	Bone scan, MRI	Female	20	Cross country running	2	Rest, diet consultation	20	Tibia (2 times), Femur- 6 months ago
10. Johnson et al ⁸	2001	Left	Ala and sacroiliac joint	Bone scan, MRI, CT	Female	21	Cross country running	2	Partial weight-bearing, diet counselling	40	Femur (2 times)- 2 and 4 years ago
11. Delvaux et al ⁹	2001	Left	Ala	Bone scan, CT	Male	28	Long distance running	1	Rest	12	None
12. Shah et al ¹⁰	2002	Left	Ala	MRI	Female	16	Volleyball	4	Rest- analgesics	4	None
13. Slipman et al ¹¹	2003	Left	Ala	MRI	Female	20	Hockey	4	Rest, analgesics (NSAIDs)	10	None
14. Silva et al ¹²	2006	Right	Ala	MRI	Male	46	Tennis	4	Rest	6	None
15. Haun et al ¹³	2007	Left	Ala	Bone scan, MRI, CT	Female	26	Long distance running	-	Rest	-	None
16. Fredericson et al ¹⁴	2007	Right	Ala	MRI	Female	21	Cross country running	-	Rest, estrogen supplementation	16	None
17. Fredericson et al ¹⁴	2007	Right	Ala	MRI	Female	18	Running	-	Rest	20	None
18. Knobloch et al ¹⁵	2007	Right	Massa lateralis	MRI	Female	22	Long distance running	2	Rest	8	None
19. Rodrigues et al ¹⁶	2009	Right	Ala	MRI, Bone scan	Female	34	Long distance running	-	Rest	36	None

Table 2. (Cont. from previous page).

Author	Year	Fracture side	Fracture-region	Imaging confirming diagnosis	Gender	Age	Type of activity	Duration of symptoms before Diagnosis (Weeks)	Treatment	Duration of symptoms after diagnosis (Weeks)	History of stress Fractures
20. Hameed et al ¹⁷	2011	Right	Ala	MRI	Female	43	Running	2	Rest, no-weight bearing , calcium, Vit D	16	None
21. Kahanov et al ¹⁸	2011	Right	Ala	MRI	Male	23	Cross country running	4	Rest, calcium, Vit D	16	None
22. Battaglia et al ¹⁹	2012	Right	Supero-medial ilium and sacroiliac joint	MRI	Female	58	Long distance running	4	Rest, analgesics (NSAIDs)	4	None
23. Tzoanos et al ²⁰	2013	Right	Ala	MRI	Male	38	Soccer	4	Rest, analgesics, Physiotherapy	12	None
24. Kendall et al ²¹	2013	Right	Sacroiliac joint	MRI	Male	25	Weightlifting	52	Rest	12	None
25. Marchinkow et al ²²	2014	Bilateral	Ala	CT, MRI	Male	20	Sprinting and throwing (javelin)	104	Rest, analgesics (NSAIDs)	20	None
26. Bednar et al ²³	2015	Right	Ala	MRI	Female	26	Horse Riding	8	Rest, analgesics (NSAIDs)	6	None
27. Takahashi et al ²⁴	2016	Left	Ala	MRI	Male	18	Rugby	7	Rest	12	None
28. Burgess et al ²⁵	2017	Left	Ala	CT, Bone scan	Male	19	Military training	2	-	-	None
29. Yuasa et al ²⁶	2017	Left	Ala	MRI	Female	16	Badminton	4	Rest	12	None
30. Baillieul et al ²⁷	2017	Bilateral	Ala	CT, MRI	Male	36	Running	104	Rest, teriparatide	24	None
31. Vajapey et al ²	2019	Right	Ala	MRI	Female	16	Running	8	Rest, analgesics Vit D, calcium	24	Tibia (bilateral)
32. Vajapey et al ²	2019	Left	Ala	MRI	Female	21	Running	16	Rest, analgesics, Partial weight-bearing, physiotherapy, Vit-D, Teriparatide (4 months after diagnosis due to delayed-union)	48	None
33. Vajapey et al ²	2019	Left	Ala	MRI	Female	22	Running	2	Rest, Vit-D, calcium	4	Tibia (bilateral, Metatarsal)
34. Vajapey et al ²	2019	Right	Ala	MRI	Male	21	Running	0.5	Rest, Partial weight-bearing, Vit D	2	Superior pubic rami (bilateral)
35. Vajapey et al ²	2019	Left	Ala	MRI	Male	53	Distance running (marathon)	2	-	-	None
36. Kavucku et al ²⁸	2020	Left	Ala	MRI	Female	42	Golf	2	Rest, analgesics (NSAIDs), Physiotherapy	16	None
37. Nomura et al ²⁹	2020	Left	Ala	MRI	Female	11	Running	0.5	Rest, partial weight-bearing	4	None
38. Ficek et al ³	2020	Right	Ala	MRI	Female	17	Tennis	6	Rest	-	None

January 2000 until January 2021, a total of 38 FSF reported cases have been located^{2,3,5-29}. The mean age of these cases was 26.2 years of age (SD=11.7), while the majority of these injuries involved females (26 out of 38 cases; 68.4%). The time between the onset of symptomatology and the diagnosis varied between 0.5 and 104 weeks, with mean time being 13.7 weeks (SD=26.6), while the mean symptoms' duration after diagnosis was 19.3 weeks (SD=15.6). Running, including long-distance and cross-country running, were the main activities associated with FSFs (21 cases; 55.3%), while history of a fatigue fracture was reported in 6 cases or 15.8%. The female gender, the young age, as well as the type of activity seem to be associated to higher risk of FSF. Details regarding fracture region, imaging confirming the diagnosis, as well as treatment may be seen in detail in Table 2.

The present 11 year retrospective study evaluated the characteristics of patients suffering FSFs, treated in the Orthopaedic Department of the "Venizeleion" general hospital in Crete, Greece. The majority of patients were females (6 patients; 75%), while running was recorded as the main causative sports activity (5 patients; 62.5%). It is of note that no patients under 18 years old were recorded in this study. However, there are many cases of bone maturity even under the age of 18 and SSFs may be observed also in adolescents. It has been documented that lower back pain in developing athletes includes SSFs in the differential diagnosis, with frequency of about 1.5%³⁰.

The main prevalence of these injuries in the female gender has already been described. In those cases, the female athlete triad, including amenorrhea, disordered eating and osteopenia should be out-ruled. Increased training intensity in young adults, as well as deficient diet, including malnutrition, low caloric or vitamin intake are also risk factors for a FSF^{1-3,14,17,22}. In the present study, although most patients were females (75%), no cases of amenorrhea were recorded. All females reported to have regular normal menstrual cycle, while no further information regarding the menstrual status were available.

It is of note that in the present study the mean duration of symptoms prior to diagnosis was 8.5 weeks, while according to the literature review this time-interval was found to be 13.7 weeks, ranging between 0.5 and 104 weeks. FSFs have an insidious clinical presentation, with low-back, pelvic or groin pain, without history of trauma^{6,31,15,16,21}. Accompanying neurological symptoms, such as radiculopathy, myelopathy, paresthesia and sphincter dysfunction may present in cases of sacral body involvement and may lead to misdiagnosis, since numerous clinical entities appear similarly^{2,31}. Physical examination may reveal sacrum tenderness, while the Gaenslen's and flexion-abduction-external rotation (FABER) test may be positive³¹. In the present case series, the main symptom was low back pain in the majority of cases (75%). The physician's awareness should be high in cases of patients participating in sports for fatigue injuries, in order to minimize the time between symptoms onset and diagnosis.

There are numerous muscles, such as gluteus maximus, piriformis, sacrospinalis, that attach to the sacrum and when

strained could have a similar clinical presentation. Likewise, ligaments, such as sacroiliac, iliolumbar, sacrospinous and sacrotuberous, present with sacrum pain if injured³¹. Sacroiliitis an inflammatory condition, as well as other clinical entities, including spondylolysis, should be kept in mind in the differential diagnosis of fatigue sacral fractures, while other clinical entities in patients engaging in sports activities, such as athletic pubalgia and femoral-acetabulum impingement should be kept in mind³²⁻³⁴.

Detailed medical history, as well as clinical examination are of utmost importance in order not to mis-or under-diagnose cases of FSF. In the present series two patients suffered in the past another fatigue fracture, while another two suffered one during follow-up. From the literature review (Table 2), about 16% of patients had sustained a fatigue fracture in the past. It seems that a fatigue fracture in any region represents a predisposing factor for such injuries in the future and this information should be evaluated during initial patient evaluation.

In the present study, most of cases (7 out of 8) were diagnosed through MRI, while in one case diagnosis was established through CT scan. Furthermore, from the literature review, as shown in Table 2, in many cases multiple imaging techniques may be used for the diagnosis of these cases. In the majority of cases MRI provided sufficient information regarding fracture characteristics. MRI was performed, alone or in combination with other imaging, in 35 of the reviewed cases (92.1%), while bone scintigraphy in 13 (34%) and CT in 10 (26%).

It seems that the preferred imaging technique is the MRI, since it has been found to be the most accurate in occult, as well as fatigue fractures^{2,35}. T1 weighted images detect FSFs as low signal intensity lines surrounded by bone marrow oedema, while in T2 weighted images they appear as high signal intensity lines. T2 weighted short tau inversion recovery (STIR) views seem to be even more sensitive in revealing the fracture. Additionally, MRI has no limitations in cases of pregnancy or lactation period^{36,37}. However, other imaging techniques may be useful for such cases, including plain X-ray view, CT scan and Bone scintigraphy.

Plain X-rays represent a screening imaging method that usually fails to demonstrate the FSF. Nevertheless, anteroposterior, as well as lateral views of the pelvis, sacrum and lumbar spine may detect complete fractures, while in some cases the fracture is apparent later on with the formation of callus^{35,38}.

Bone scintigraphy with technetium 99^m medronate methylene diphosphonate is a sensitive method for revealing fractures. The main disadvantage, besides the low specificity, is that due to the fact that sacroiliac joints have a high uptake of medronate methylene diphosphonate, bilateral sacral fractures cannot be diagnosed^{35,38}.

Computer tomography (CT) is a valid imaging technique for detecting a FSF, since it is both sensitive and specific. In some cases, it may be used in combination with magnetic resonance imaging (MRI) for ruling out malignancy and infection^{1,38}.

Regarding treatment of FSFs, cessation of high-impact activities for at least 6 weeks is proposed^{1,2}. All the presented patients had rest, while in some patients partial weight-bearing with crutches was implemented. Furthermore, vitamin D and calcium were given in 3 cases. Calcium and vitamin D supplementation is also proposed for such cases². The need for and doses of supplements vary dependent on the athlete's diet and season of the year.

All presented patients received analgesics, without the use of non-steroid anti-inflammatory drugs, while two of them were supported with physiotherapy. Avoidance of anti-inflammatory medications may be considered given their potential to delay bone healing, while physiotherapy for core strengthening and stretching may help the athlete's range of motion and minimize the need for retraining when returning to sports^{1,38}.

It is of note that one case presented with delayed union (case 4 in Table 1). The patient was commenced on teriparatide for 24 weeks, having successful union. From the reviewed cases, the same treatment received two patients with delayed union (cases 30 and 32 from Table 2)^{2,27}. Hence, it seems that teriparatide should be considered in such cases.

Although, surgical treatment of these injuries is not considered the first line management, it is recommended in cases of neurological deficit, instability and severe sacrum alignment disruptions. Surgical management includes internal osteosynthesis with the use of screws or hinge fixation^{38,39}. None of the presented or the reviewed FSF cases underwent surgical treatment.

The present study has some limitations. It is a retrospective one, while the sample is relative small, coming from a single center. Nevertheless, taking into account the existing literature, it represents the largest series, while it provides a thorough literature review with all reported FSF cases, covering 21 years from 2000 until 2021.

In conclusion, FSFs represent a clinical entity often misdiagnosed. Hence, the differential diagnosis for chronic low back or posterior hip pain in young patients engaging in sports, especially female runners, should include sacral fatigue fracture. History of other fatigue injuries seems to be a predisposing factor. It is of utmost importance to obtain advanced imaging, including MRI, early in the workup for identifying a sacral fatigue fracture and preventing worsening of the injury.

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