

A Case of Parasympyseal and Associated Insufficiency Fractures of Pubic Rami in a Patient with Mixed Connective Tissue Disease

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Parasympyseal insufficiency fractures are uncommon. Furthermore, none have been reported in systemic rheumatic diseases other than rheumatoid arthritis. In this article we report on parasympyseal insufficiency fractures in a patient with mixed connective tissue disease.

Key Words : *Insufficiency fracture ; Parasympyseal ; Mixed connective tissue disease*

INTRODUCTION

Parasympyseal insufficiency fractures (PIF) are not a new disease but have been recently recognized, with the first report being published by Goergen et al. in 1978¹⁾. To date, details of about 65 cases have been reported^{2-11,14,15)}. Among these cases, only 14 cases were associated with rheumatoid arthritis^{2-5,9,14)}. To the best of our knowledge, however, no cases of PIF from other systemic rheumatic diseases other than RA have been published. We report on a patient with mixed connective tissue disease (MCTD) who developed parasympyseal and associated insufficiency fractures.

CASE

A 39-year-old woman was admitted to the hospital because of left groin pain. She had a past medical history of meningitis. Four years previously, she was found to

have MCTD on the clinical diagnostic criteria of Sharp et al. At that time, she experienced arthritis, Raynaud's phenomenon, swollen hands and biopsy-proven myositis. Laboratory examinations revealed the following results: leukocyte count 3600/mm³, antinuclear antibody 1:640, speckled pattern, positive anti-nRNP & Ro antibody, LDH 208 U, CK 23 U. The patient was treated with hydroxychloroquine, prednisolone (10 to 20 mg/day, sometimes on 5 mg/day), piroxicam and angiotensin-converting enzyme inhibitor or calcium channel blocker. Thereafter, she was intermittently admitted to the hospital due to drug-induced hepatitis, severe dysplasia of the uterine cervix, fever with chills and painful lymphadenopathy. At the presentation, she visited the hospital due to back and left groin pain during walking down stairs about 15 days ago. She denied a significant trauma history. Pelvis X-ray previously checked was unremarkable except for an osteoporotic change (Figure 1A). Physical examination on admission showed positive a Patric test on the left hip joint. We suspected her to have the avascular necrosis of the left femoral head. However, MRI of the left femoral head did not confirm our suspicions but instead, revealed a synovitis with effusion. Simple pelvis radiograph was done. A plain radiograph of the pelvis revealed insufficiency fractures of parasympyseal and left pubic rami

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Figure 1. Insufficiency fractures: MCTD in 40 year-old woman.

- (A) An anteroposterior view of plain radiograph of the pelvis taken on Nov. 1995 shows osteoporosis.
- (B) She developed left groin pain aggravated by activity. Follow-up plain pelvis study on Jan. 1997 reveals fractures (arrows) in the right parasymphyseal bone and left pubic rami, accompanied by osteolysis, osteosclerosis and bone fragmentation.
- (C) Five months later, healing of the fractures with callus formation is noted.

(Figure 1B). We recommended bed rest, tapering of prednisolone and management of steroid-induced osteoporosis. After 5 months, a follow-up pelvis X-ray revealed the healing of parasymphyseal fracture (Figure 1C).

DISCUSSION

Stress fractures are classified as insufficiency and fatigue fractures. Insufficiency fractures are a form of stress fracture, which occur when normal or physiological

muscular stress is applied to the bone with deficient elastic resistance or mineral content^{2,8)}. An uncommon site for such a fracture is through the body of the pubis adjacent to the symphysis, hence the term parasymphyseal. To date, details of about 65 cases with PIF have been reported. However, PIF associated with other systemic rheumatic disease other than rheumatoid arthritis have not been published. Only 14 patients with rheumatoid arthritis were associated with PIF (Table 1). Among these patients, only 2 had previously undergone total hip arthroplasty and the others denied significant trauma history. The diagnosis is almost exclusively made in postmenopausal women with osteopenic bone. Postmenopausal osteoporosis, rheumatoid arthritis and steroid therapy, previous hip replacement and pelvic radiotherapy are represented as etiologic factors of PIF³⁻⁸⁾.

The specific ring-like anatomy of the pelvis and the forces acting on this area, such as external rotation, lateral compression, vertical shear, or their combination, during trauma or repetitive abnormal muscular stress may be responsible for the occurrence of more than one pelvic fracture. The sacral fractures may precede or occur simultaneously with the pubic fractures²⁾, although chronology may be indeterminable. So the concomitant existence of parasymphyseal and other, frequently occult, insufficiency fractures has recently been emphasized. As a result of parasymphyseal insufficiency fractures, lysis and callus formation produce a destructive, malignant-appearing lesion³⁾. Indeed, a malignancy with a pathologic fracture was considered in a patient with rheumatoid arthritis and concomitant parasymphyseal insufficiency fracture in our hospital, which was later proved as hematoma by sono-guided aspiration biopsy. A biopsy of these lesions shows histologic evidence of bone damage and repairing tissues. The histologic changes consist of various quantities of hemorrhage, fibrosis, woven bone and cartilage formation (callus), and even fragments of dead bone. Inflammation is minimal to absent. The radiologist and pathologist should be aware of this entity, so inappropriate therapy, including en bloc resections, is not to be done.

Plain film radiograph is usually sufficient for evaluation of the pubic fractures. Radionuclide bone scanning is sensitive for fracture detection but lacks specificity and need not be done routinely if the plain films and tomograms are typical for insufficiency fracture^{2-5,8-10)}. Although not usually necessary, computerized tomography is believed to be the most accurate means for

Table 1. Parasymphseal insufficiency fractures in patients with rheumatoid arthritis

Author	Number of patients	Duration of RA	Sex / Age	Site of PIFs	History of Trauma	Associated fracture
Davies AM ²	5	ND	F/55 78	ND	ND (2-THA)	Present, but ND
Casey D ³	3	19	F/59	Right	Strenuous activity	(-)
		ND	F/50	Bilateral	No	(-)
		15	F/66	Bilateral	No	Left acetabulum insuff Fx
Tauber C ⁴	1	16	F/40	Symphysis disruption	No	Stress Fx of Left ilium Fatigue Fx of Left Superior pubic ramus after THA
Arafat QW ⁵	1	12	F/62	Left	No	Bilateral SIFs
Cooper KL ⁹	1	ND	F/71	Left	No	Bilateral SIFs, Left Superior acetabulum insuff Fx
McGuigan LE ¹⁴	1	23	F/76	Left	No	(-)
Peh WCG ¹⁵	2	ND	F/72	Bilateral	No	Left SIF
		ND	F/72	Right	No	Bilateral SIFs

RA; rheumatoid arthritis, PIFs; parasymphseal insufficiency fractures, ND; not documented, THA; total hip arthroplasty, Insuff; insufficiency, Fx; fracture, SIFs; sacral insufficiency fractures

demonstrating insufficiency fractures and for ruling out malignancy^{2, 10}). Recently given the prevalence of MR units, a MRI of pubic parasymphseal insufficiency fractures is used and characteristically demonstrates a hyperintense mass lesion with a hypointense rim on T2-weighted imaging, showing peripheral and septal enhancement after contrast administration⁹). Therefore, MRI is helpful to distinguish fractures from the bone tumor.

The treatment of pelvic insufficiency fracture is generally conservative¹⁵). It should be directed at pain relief and limitation of weight bearing activities. When osteoporosis is the etiological factor, the plan of management should also include osteoporosis therapy.

There are some cases with parasymphseal fractures in rheumatoid arthritis patients which were published in English literature. Our case emphasizes that systemic rheumatic diseases, requiring long-term low-dose steroid use as well as RA, may be a predisposing factor of parasymphseal fractures. Conclusively, the possibility of a pelvic fracture should be considered in patients with systemic rheumatic disease, as well as RA presenting with spontaneous low back and inguinal pain, and investigation should include serial plain films and a bone scan.

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