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ORIGINAL STUDY

Osteonecrosis in the lunate bone in the presence of palmar lunate dislocation in rheumatic patients

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This study aims to assess the relationship between rheumatic disorders with palmar lunate dislocations and lunate osteonecrosis (ON). From 2004 to 2016, we prospectively studied 24 wrists with advanced rheumatic disease in which the lunate was chronically dislocated palmarly, situation that clearly favors the apparition of ON. There were 18 female and 6 male with a mean age 64 (52-79) years old. The surgical treatment was wrist arthrodesis in 19 cases and wrist arthroplasty in 5. We evaluated X- ray data and we correlated preop X-ray, blood test parameters and MRI data with histopathological findings. Statistical analysis of data was performed with Chi square test and Student´s t-test (significance level $p \leq 0.05$). We found a focal lunate necrosis in two cases. We did not find a statistically significant correlation between ON and lunate dislocation (p= 0.793). We did not find any blood test parameter statistically significant associated with ON (p= 0.621). Despite the possibility to developing ON of the lunate bone in presence of palmar lunate dislocation in patients with rheumatic wrist, angiogenesis and synovitis formed could have a role in chronic adaptation to the loss of vessels in the lunate.

Keywords : angiogenesis ; Kienböck´s disease ; lunate histology ; osteonecrosis ; rheumatoid disorders.

INTRODUCTION

Osteonecrosis (ON) is an entity characterized by death of bone marrow and trabecular bone as a result of a compromised arterial supply. It may follow a trauma resulting in the displacement of a bone fragment that disrupts blood supply causing bone ischemia (post-traumatic ON) or may complicate the course of many systemic diseases or conditions (atraumatic ON) *(5)*. Systemic conditions known as risk factors for ON are corticosteroid treatment, sickle cell disease, thrombophilia, dyslipemia, alcoholism and chronic inflammatory diseases. Rheumatoid diseases such as Systemic Lupus Ery-

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thematosus (SLE) and Rheumatoid Arthritis (RA) are frequently associated with systemic bone loss and ON *(8,9,25)*. In advanced rheumatic disease in which the lunate was chronically dislocated palmarly it is a situation that clearly favors the apparition of ON. Mok et al. *(14)* identified a patient that developed lunate ON during the course of RA in the absence of steroid treatment. Desy et al. *(7)* reported a case of Kienböck´s disease and juvenile idiopathic arthritis. Arner et al.*(2)* identified a patient with complete palmar lunate dislocation in RA and reported avascularity without avascular changes. Usually, in a case of lunate necrosis a pathological examination of the excised bone showed extensive necrosis, collapse, and invasion by vascular fibrous tissue. In the wake of these changes, a secondary degenerative arthritis was developing *(16,20)*. MRI has become the best technique for early recognition of ON considering the high sensibility and specificity. In cases of ON, these cells die and the signal lost. Ischemic necrosis results in a decreased signal on T1-weighted imaging *(21,23,24)*.

The purpose of this study was : 1) to describe the presence of lunate ON in rheumatic disorders associated with palmar lunate dislocation ; 2) to investigate the correlation between the histology with MRI, radiological and blood test parameters.

MATERIALS AND METHODS

A prospective case series study from January 2004 to January 2016 was performed. There were 24 patients that were operated for rheumatoid disorders in the wrist. There were 16 RA, 1 SLE and 7 psoriatic arthritis (PA). Eighteen patients were female and six male with a mean age 64 (range, 53 to 79 years). In all the cases the lunate was chronically dislocated palmarly.

Blood was collected for analysis of the following parameters within one week before surgery : erythrocite sedimentation rate (ESR), C-reactive protein (CRP), rheumatoid factor (RF) and anti-citrullinated protein antibodies (ACPAs). Normal levels were ESR 1-10 mm/hour, CRP 0-5 mgr./L, RF < 20 UI/ml, and ACPAs < 5.3 UI/ml.

Posteroanterior and lateral radiographs were obtained to identify lunate dislocation, fracture line, lunate collapse, ulnar translation, and carpal collapse (Fig. 1). We evaluated X-ray data such as Stahl Index, Carpal Height Ratio (CHR), Carpal Ulnar Distance Ratio (CUDR) and radio-scaphoid angle. In order to determine whether compression was present in the lunate the index reported by Stahl was applied *(22)*. The height was determined by measuring the distance between the proximal and distal articular surfaces on the lateral projection, and the horizontal diameter by measuring the greatest dorso-palmar diameter perpendicular to the vertical height (Index range of 50-70). We evaluated the carpal collapse and the ulnar translation by McMurthy and Youm´s classification *(26)*. The normal CHR was 0.54 +- 0.03 mm and the normal CUDR was 0.30 +- 0.03. The normal radiocarpal angle was 47º. The wrist was evaluated preoperatively with MRI using a 1.5 Tesla Siemens vision unit. The examination was done with the following sequences : Spin-echo pulse sequences (SE) SET1 and SET2. T1-weighted sequences were obtained using short repetition (TR), echo delay (TE) times (TR= 600 msec, TE= 38 msec) and fat-suppressed. The carpus was imaged in transverse, coronal, and sagittal planes. T1-weighted images were evaluated for bone necrosis of the lunate.

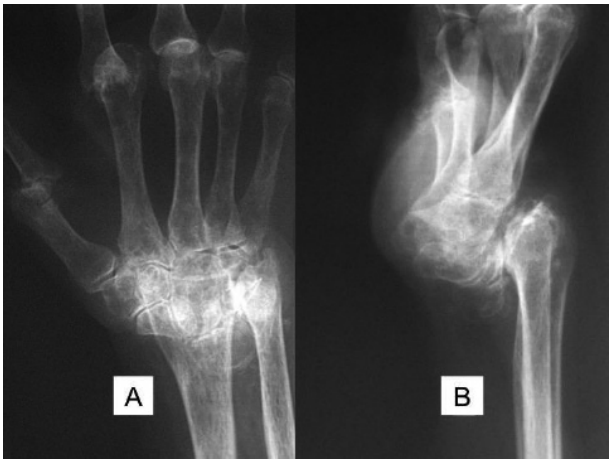

Figure 1. — Rheumatoid arthritis : A) PA and B) lateral X-ray demonstrating the palmar dislocation of the lunate and joint destruction.

We performed total wrist arthroplasty (Universal 2, Integra LifeSciences, Plainsboro, NJ) in 5 cases and wrist arthrodesis (3.5 LCP wrist fusion system, DePuy Synthes, Oberdorf, Switzerland) in the

Figure 2. **—** Lunate bone : Macroscopical appearance of a well-vascularized lunate.

other 19. In the operating room we excised whole lunates and we studied the macroscopical aspect inside and outside. We evaluated the size, cartilage damage in the surfaces, and ligament insertions. We cut the lunate bone and we evaluated the bleeding bone and collapse areas. Histopathological analysis was performed with lunate sections (Fig. 2). The lunates biopsies were fixed in formalin, decalcified, embedded in paraffin and cut in 5 μm. sections and stained with haematoxylin and eosin. We visualized lunate sections by microscope. We correlated blood test parameters with the histology. We correlated the parameters in the preop X-ray and MRI data with histopathological findings.

Data were analysed using SPSS software, version 21 (Chicago, IL, USA). For comparison between 2 groups, Chi square test or Fisher exact test was used to compare categorical measurements. Student´s t-test or Mann-Whitney U-test was used to compare continuous variables. Statistical significance was considered as p less than 0.05.

RESULTS

Microscopic analysis showed synovial hyperplasia in all the cases, fatty marrow and fibrovascular tissue in 91% (Fig. 3). Osteoarthritis was present in all the cases. We did not find ON in any complete lunate but we found a focal necrosis (empty lacunae) in two cases (Fig. 4). In 22 cases lunate histology showed fatty bone marrow, normal trabeculae and blood vessels.

Mean CRP 16.20 mg/L (range, 1-54.2), mean ESR 21.40 mm/hour (range, 5-75), mean anti-CCP

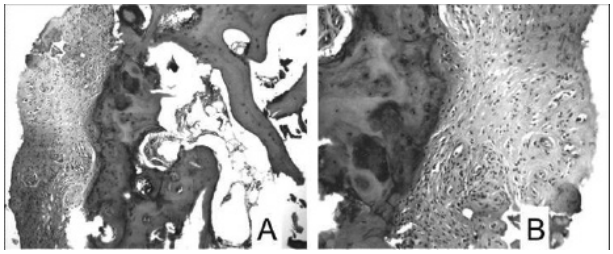

Figure 3. **—** Histopathological specimen of the lunate showing synovial hyperplasia and a reaction with numerous cells visualised in the inset under polarized light : A) Hematoxylin-eosin (HE) magnification x 100 ; B) HE x 200.

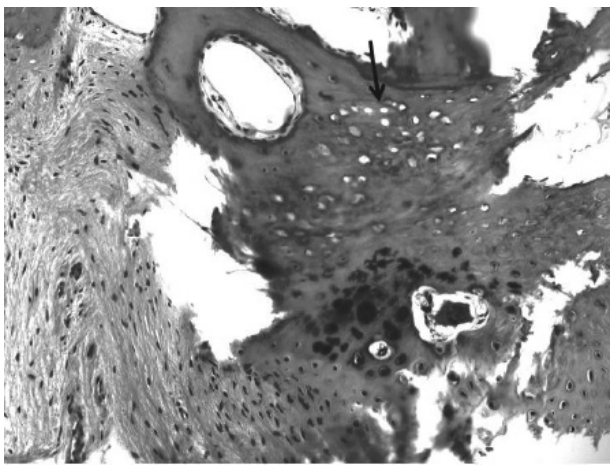

Figure 4. **—** Hematoxylin-eosin stains of an osteonecrosis demonstrating empty lacunae (signifying bone death : black arrow) and a generalized lack of hematopoietic cells (HE magnification x 200).

102.17 UI/ml (range, 1-340), and mean RF 208.24 UI/ml (range, 5-669).

We did not find any blood test parameter statistically significant associated with focal lunate necrosis (p= 0.621). ANA was positive at low title in 6 RA and at high title in 2. RF was positive in all RA.

A lateral X-ray of the wrist revealed palmar dislocation of the lunate bone in all the cases. Mean CHR was 0.375 (range, 0.25-0.45), mean CUDR was 0.301 (range, 0.20-0.38), mean Stahl Index was 57.95 (range, 30.7-71.0), and the mean radio-scaphoid angle was 74º (range, 68º-82º). We found a greater ulnar translation in RA than in other rheumatoid disorders (CUDR Student-T-test p = 0.002). Radiology of the wrist demostrated erosive changes. We did not observe any correlation

between ON in the histology and palmar lunate dislocation (p= 0.793). In all the cases MRI confirmed the presence of synovitis, osteoarthritis and joint destruction. In two cases we found a lunate flattening associated with a partial hypointensity in coronal sections in T1-weighted images in MRI. We did not find a statistically significant correlation between lunate ON and scapholunate ligament rupture (Chi-square of Pearson p= 0.640). We did not observe any correlation between ON in the histology and the differents sections in MRI SE T1/ T2/ fat suppression/STIR (Chi-square of Pearson p= 0.480 ; p= 0.699, p= 0.815, p=0.773).

DISCUSSION

Osteoarthritis and ON are arthropathies that result in marked pain and disability *(19)*. ON and its association with rheumatoid disorders is extremely rare. Some authors showed a relationship between ON and rheumatoid disorders *(7,12,14)*. It is known that ON is associated with the intake of corticosteroids *(13,14,27)*. However we do not know the role that chronic dislocation of the lunate plays in the appearance of ON *(2,3)*. In spite of the severe ligamentous destruction in RA, not always we can detect ON of the lunate although it exists, by radiographs and MRI. In our study, we did not find statistically significant correlation between ON in the histology and palmar lunate dislocation. This suggests that ON in the lunate bone in rheumatic disorders has another main cause not the lunate chronic dislocation. On the other hand, although there is necrosis this is partial not complete with some capacity for regeneration. In the histological study by Hashizume *et al. (10,11)* necrotic areas were invaded with new bone formation and granulation. The necrotic tissue then changed into fibrous scar tissue and necrotic debris. Around the necrosis area, non-necrotic tissues under hypervascularized conditions were reactive to the necrosis.

Clinical factors that have been identified as predictors of poor outcomes include RF positivity and evidence of active inflammation (elevated ESR and CRP level) *(4,15,18,27)*. In the study by Amos et al. *(1)* CRP levels and ESR were measured in 56 patients with RA. Radiographic damage was significantly more likely to occur when serum CRP and ESR were persistently raised. In our study we did not find any blood test parameter statistically significant associated with ON.

In our study MRI confirmed the presence of synovitis, osteoarthritis and joint destruction in all the cases. In two cases we found a lunate flattening associated with a partial hypointensity in coronal sections in T1-weighted images in MRI suggestive of partial necrosis. In normal bone, T1-weighted images MRI have a high signal and T2-weighted images show and intermediate signal. In early stages of ON, T1-weighted images exhibit a low signal and T2-weighted images show a high signal *(17,21)*. Desser *et al. (6)* found that MRI was able to distinguish areas of viable and nonviable bone within the lunate. Trumble and Irving found a correlation between the loss of signal intensity on T1 and T2-weighted MRI images and evidence of ON by histology *(23,24)*. Our results support the hypothesis that there are other causes, not only the palmar lunate dislocation that produce the lunate necrosis in rheumatic patients. This study suggests that there is a process of repair, angiogenesis and chronic adaptation to the loss of vessels that prevents complete necrosis.

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