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Rheumatology

# Ankylosing Spondylitis and Autoimmune Thrombocytopenia: the First Senegalese Case

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Abstract Case Report

Introduction: Ankylosing spondylitis (AS) and autoimmune thrombocytopenia are two distinct diseases, and they can occur rarely. Objective: To determine the epidemiological, clinical, therapeutic profiles and impact of this rare association in the literature Observation: This was a 57-year-old teaching patient received on September 07, 2020. The examination of the musculoskeletal system revealed lumbar pain. The onset was 26 years ago and was marked by the progressive onset of pain located in the dorsal spine, lumbar and manubrio-sternal region, then a progressive extension to both wrists and the left knee with a VAS of 6/10. Associated signs were stiffness and synovitis of the left knee. The biology showed a non-specific biological inflammatory syndrome with accelerated blood pressure at 70 minutes in the first hour and CRP increased to 15 mg/l, normocytic normochromic anemia (hemoglobin 7.6 g/dl), thrombocytopenia at 51,000/mm3 (N: leukopenia at 3,100/mm3 and neutropenia at 1115/mm3 (pancytopenia). The transaminases ALAT was 60 IU/L and Gamma GT was increased to 265 IU/L, the immunological tests were negative with the presence of the HLA B27 positive antigen, and the fixed anti-platelet antibodies showed the presence of fixed autoantibodies directed against the glycoprotein complex IIb and IIIa and the absence of circulating antibodies directed against the glycoprotein complexes IaIIb, IIbIIIa and IbIX. The lumbar CT scan shows syndesmophytes and osteophytes (Fig 2) and the CT scan of the pelvis shows Forestier stage III sacroiliitis (Fig 1). Conclusion: The association of ankylosing spondylitis and autoimmune thrombocytopenia is rare but possible, and should be considered whenever a known spondyloarthritis patient presents with a platelet collapse. Treatment with methotrexate and anti-inflammatory drugs may be effective in some cases.

Keywords: Ankylosing spondylitis, thrombocytopenia, autoimmune, Senegalese.

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## Introduction

Ankylosing spondylitis or rheumatic pelvispondylitis is a chronic inflammatory rheumatism characterized by a predominantly axial localization in the spine and sacroiliac joints, an involvement of the entheses and a tendency to bone ankylosis by ossification of these entheses [1-3].

It is the leading spondyloarthritis and occurs particularly in young adult males. It is the second most common chronic inflammatory rheumatic disease after rheumatoid arthritis. Autoimmune thrombocytopenia is a rare autoimmune disease that associates peripheral destruction of platelets and insufficient bone marrow production. It is increasingly well understood that it involves a humoral immune response in which B

lymphocytes, via T lymphocyte cooperation, especially with splenic follicular helper T lymphocytes, differentiate into plasma cells that produce anti-platelet antibodies, leading to phagocytosis of platelets by splenic macrophages [4-7]. Although this association has been reported in the literature, only three cases have been described, and the authors report three observations of the concomitant appearance of these two conditions. Secondly, they discuss the possibility of a common infectious triggering factor. The incidence of this association has not been reported in the literature. We report a new case of a patient followed for ankylosing spondylitis who presents autoimmune thrombocytopenia followed since 2019 in the hematology department

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### **CLINICAL CASE**

This was a 57-year-old teaching patient received in the department on September 07, 2020. On clinical examination: general examination satisfactory general condition BP was 130/70mmhg, Fc was 84b/min. examination of the musculoskeletal system finds.

Lumbar pain. With a beginning that would go back to 1995 marked by the progressive occurrence of pain located at the level of the dorsal rachis, lumbar and manubrio-sternal fixed and then progressive extension at the level of the two wrists and the left knee of inflammatory schedule calmed by the taking of anti-inflammatory of type of burn and intensity EVA 6/10 associated sign stiffness, bilateral talalgia and synovitis of the left knee no dry syndrome and no skin rash.

Complementary biology examination shows a non-specific biological inflammatory syndrome with accelerated blood pressure at 70 min at the first hour and increased CRP at 15 min, normochromic normocytic anemia (hemoglobin 7.6 g/dl), thrombocytopenia at 51,000 mm3, leukopenia at 3. 100/mm3 and neutropenia at 1115/mm3 (pancytopenia) Transaminases ALAT was 60IU/L and Gamma GT was

increased to 265IU/L Immunological tests were negative with the presence of HLA B27 positive antigen Fixed anti-platelet antibody showed the presence of fixed autoantibodies directed against glycoprotein complex IIb and IIIa and the absence of circulating antibodies directed against glycoprotein complexes IaIIb, IIbIIIa and IbIX The lumbar CT scan shows syndesmophytes and osteophytes (Fig 2) and the CT scan of the pelvis shows Forestier stage III sacroiliitis (Fig 1).

Following the clinical and biological examination and by the AMOR criteria, the diagnosis of HLA B 27 positive SpA [8], associated with autoimmune thrombocytopenia, was made in the presence of pancytopenia with a platelet count of 51,000/mm3, the presence of fixed autoantibodies directed against the glycoprotein complex and the absence of circulating antibodies directed against the glycoprotein complexes.

The treatment received was based on hydroxychloroquine 200mg 1cp 2 times/day, prednisone 5m 1cp 2 times/day and tramadol 50mg 1cp 3 times/day if pain.



Figure 1: CT scan of the pelvis showing Forestier stage III sacroiliitis



 $Figure \ 2: Lumbar \ CT \ scan \ showing \ \overline{T11, T12 \ L1} \ syndes mophytes \ and \ L4, L3, L2, L1, T12, T11, T10 \ osteophytes \ with \ disc \ voids$ 

#### **DISCUSSION**

Discussion Our observation is that of an HLA B27 positive ankylosing spondylitis associated with autoimmune thrombocytopenia. Our case is of epidemiological, diagnostic, prognostic and therapeutic interest.

Epidemiologically, the exact prevalence has not been reported 3 cases reported in the literature. In Europe, 2 cases including 1 case in the department of internal medicine at the University Hospital Ranguel Toulouse reported in 1990 by D. Schlaifer *et al.*, [9], one case in 1993 reported by B. Pallot-Prades *et al.*, in the department of internal medicine at the University Hospital Bellevue Toulouse [10] and one case which differs from the two other cases because of associated chronic thyroiditis was reported in Japan in 1992 by K. Uozumi *et al.*, in the department of internal medicine at the University Hospital Bellevue Toulouse [11]. Uozumi et all in the department of internal medicine of Kagoshima University Hospital [11].

In America no case has been reported, in Africa more particularly in Senegal was the 1st case in our department. The incidence has not been reported in the literature, according to the 3 cases reported the average age is between 43 and 58 years with a male predominance of 2 men on one woman according to the 3 cases reported. Our patient is male, 57 years old.

The diagnosis of ankylosing spondylitis was made clinically with the presence of a pelvic-spinal syndrome of lumbar predominance with manubrium-sternal pain associated with bilateral talalgia and asymmetrical polyarthritis in the two wrists and the left knee.

At the paraclinic, we have a SIBNS VS and CRP very increased FR negative, antinuclear antibody negative, Ag HLA B27 positive at the CT of the pelvis we have a sacroiliitis stage III of Forestier helped by the criterion of AMOR which gave us a Score 13pt>6.

Autoimmune thrombocytopenia was diagnosed in front of pancytopenia with a platelet count that was 51.000/mm3, the presence of fixed autoantibodies directed against the glycoprotein complex and the absence of circulating antibodies directed against the glycoprotein complexes. B. Pallot-Prades et all in their case reported in the literature hospitalization in rheumatology is motivated by the appearance of an inflammatory pain of the interphalangeal of the big toe, a hydarthrosis of the left knee the patient feels the pain of the dorso-lumbar junction [10]. The radiographic examination of the knees and feet was normal, but there were syndesmophytes of the dorsal-lumbar junction, the images were found on an unprepared abdominal film. The sacroiliac X-rays were normal, and biologically, we noted a very regenerative normocytic anemia at 7.7 g/dl, thrombocytopenia at 10,000/mm3 and numerous

schizocytes, a nonspecific biological inflammatory syndrome with increased SV and CRP, and a very low level of inflammation. Uozumi et all in their case reported in the literature, the patient had dorso-lumbar pain with the presence of a bamboo spine on radiology, a non-staging sacroiliitis, a monoarthritis of the right knee, an idiopathic thrombocytopenia associated with chronic thyroiditis and the presence of the HLA B27 antigen which was not the case of our patient. On the other hand K. Uozumi et all in their case reported in the literature, the patient had dorso-lumbar pain with the presence of a bamboo spine on radiology, non-staging sacroiliitis, monoarthritis of the right knee, idiopathic thrombocytopenia associated with chronic thyroiditis and the presence of HLA B27 antigen which was not the case of our patient [11].

In our patient he was submitted to hydroxychloroquine 200mg 2 times/day, prednisone 5m 2 times/day and tramadol 50mg 1cp/day.

## **CONCLUSION**

Ankylosing spondylitis is chronic inflammatory rheumatism, which belongs to the group of spondyloarthritides, characterized by a predominant axial localization in the spine and sacroiliac joints, by the involvement of the entheses and by a tendency to bone ankylosis. Autoimmune thrombocytopenia is a rare autoimmune disease that associates peripheral destruction of platelets and an insufficiency of medullar production. The association of ankylosing spondylitis and autoimmune thrombocytopenia is rare but possible and should be considered whenever a known spondyloarthritis patient presents with platelet collapse. Treatment with methotrexate and anti-inflammatory drugs may be effective in some cases.

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