Case Report
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Sheehan syndrome announcing rheumatoid arthritis: a case report

Abstract

Occurrence of arthritis and rheumatism in patient with Sheehan syndrome seems to be significantly higher than that in the general population. The association with an authentic rheumatoid arthritis (RA) remains however exceptional and unusual. The chronology of the association is variable, but the concomitant occurrence of these two diseases remains exceptional: to the best of our knowledge, only three observations of synchronous occurrence of RA and Sheehan syndrome have been found in the medical literature. We report an original observation of concomitant onset of RA and Sheehan syndrome occurring after normal delivery in a 43-year-old Tunisian woman.

Keywords: sheehan syndrome, rheumatoid arthritis, hypophysitis, empty sella

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Introduction

Specific endocrine damage is exceptional during rheumatoid arthritis (RA).[1,2] These are mainly quantitative and/or qualitative abnormalities of the gonadal, adrenal, prolactin, and thyroid hormones. These hormonal abnormalities have a demonstrated effect on the incidence and activity of this rheumatic disease.[3,4]

The involvement of the pituitary gland during RA is exceptional.[5,6] It often results from lymphocytic[5] or xanthomatous[6] hypophysitis accompanying RA and which can result in a partial or pan-hypopituitarism. The inaugural forms are unusual and represent a real diagnostic challenge.[5] We report an original observation of concomitant onset of RA and Sheehan syndrome occurring after normal delivery.

Case report

43-year-old Tunisian woman, with no pathological medical history, was explored for marked asthenia with inflammatory polyarthralgia occurring one week after normal delivery. The somatic examination noted hypotension at 90/58, tachycardia at 110/min, and marked signs of extra cellular dehydration. No fever, lymphadenopathy, active skin lesions, or synovitis have been noted. Biology showed hypoglycemia at 3.6mmol/l, hyponatremia at 128mmol/l, and erythrocyte sedimentation rate at 48mmH1. The infectious investigations were negative. The hormonal tests objectified a central hypothyroidism with Thyroid Stimulating Hormone (TSH) at 1.3µIU/ml and total free thyroxin (FT4) at 2pmol/l and central adrenal insufficiency with cortisol at 42ng/l and positive short synacthen test. The other hormonal dosages were normal. The immunological tests were negative (anti-thyroglobulin, anti-thyroperoxidase, anti-nuclear, anti-neutrophil cytoplasm, and anti-native DNA antibodies). Painful joint radiographs showed no abnormalities. The pituitary magnetic resonance imaging objectified an extinction of the pituitary gland with an empty sella.

The diagnosis of Sheehan syndrome was retained. Polyarthralgia was considered to be a possible joint manifestation of this syndrome. The patient was treated with levothyroxine in progressive doses up to 150µg/day and hydrocortisone at a dose of 30mg/day with a favorable outcome: disappearance of asthenia, normalization of blood pressure, blood sugar, and serum ionogram.

Six months later, the patient returns to our consultation for persistent inflammatory arthralgia of the hands, wrists, and feet, which became very debilitating with significant morning stiffness. The somatic examination noted synovitis of the two wrists, metacarpo-phalangeal joints, and proximal inter-phalangeal joints of both hands. Her Sheehan syndrome was well managed with normal TSH, blood sugar and serum ionogram. Biology showed a marked biological inflammatory syndrome with inflammatory anemia at 10g/dl, erythrocyte sedimentation rate at 96mmH1, and C-reactive protein at 22mg/l. Radiology objectified multiple erosions on hands and forefoot radiographs. The rest of the basic bioassays were within normal limits: leukocytes, platelets, calcemia, uric acid, transaminases, muscle enzymes, lipid parameters, and serum protein electrophoresis.

The immunological assessment showed: positive latex test at 256IU, positive Waaler-Rose reaction, negative anti-nuclear antibodies, negative anti-native DNA antibodies, and positive anti-citrullinated protein antibodies (anti-CCP antibodies) at 57IU. The diagnosis of rheumatoid arthritis was retained. The assessment of systemic manifestations was negative. The patient was treated with prednisone, hydroxychloroquine, and methotrexate with favorable outcome. This RA was already concomitant with Sheehan syndrome but was initially undiagnosed.

Discussion

Describe first in 1937 by Sheehan HL as an anterior pituitary insufficiency due to ischemic pituitary necrosis complicating severe postpartum hemorrhage,[7] Sheehan syndrome remains a significant cause of hypopituitarism in developing countries.[8,9] However, it remains under diagnosed and sometimes under treated due to the variable and often latent nature of hormonal deficits.[10] It can exceptionally occur, as for our patient, after normal delivery without massive bleeding or hemorrhagic shock.[8–10] Occurrence of arthritis and rheumatism seems to be significantly higher than that in the general population:[11,12] 13.5% versus only 5.9% in Sheehan HL et

al.[11] series. The association with an authentic RA remains however exceptional and unusual. Indeed only a few sporadic observations have been reported in the world literature.[5,6,12]

The chronology of the association is variable[5,6,12] but the concomitant occurrence of these two diseases remains exceptional: to the best of our knowledge, only three observations of synchronous occurrence of RA and Sheehan syndrome have been found in the medical literature.[12] The hypopituitarism associated with or complicating RA seems to be of autoimmune origin (autoimmune hypophysitis with pituitary extinction and empty sella).[5,6,12] These hormonal dysfunctions are important to diagnose and treat properly because they contribute to the development, persistence, and severity of RA.[3,4]

Conclusion

Rheumatic manifestations are common in women with Sheehan syndrome, but the association with authentic RA remains exceptional and unusual. This association seems to be of autoimmune mechanism (autoimmune hypophysitis), and is characterized by marked severity of rheumatic disease. So it seems useful to screen for RA, women newly diagnosed Sheehan syndrome, especially if there is no improvement or persistence of joint complaints after treatment for hormonal deficits. Our observation is distinguished by the occurrence of Sheehan syndrome after normal delivery, and by the concomitant onset of RA and Sheehan syndrome.

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Conflicts of interest

The authors declare there are no conflicts of interest.

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