Advances in
Clinical and Medical Sciences
Volume 2, Issue 2

Case Report

Is it Coincidence of Sacroiliitis and a Mild Form of Idiopathic Inflammatory Myopathies? A Case Report

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Abstract

Sacroiliitis is an aching inflammation of the specific sacroiliac joint which often presents with lower back pain. The idiopathic inflammatory myopathies, also called myositis, are a heterogeneous group of diseases which contribute to symptoms of muscle weakness and inflammation. It is reported that sacroiliitis with myositis can occur because of infections, malignancy, or rheumatoid diseases. We present a case of 57-year-old male experiencing lower back pain. There were abnormal findings in antibody examination test, wholebody bone scan, and quantitative sacroiliac scan. As a result, he was then diagnosed with sacroiliitis and mild form of idiopathic inflammatory myopathies. We discovered that even without underlying disease which we mentioned above, there is still a correlation observed in sacroiliitis and mild form of idiopathic inflammatory myopathies.

Keywords: Sacroiliitis, Myositis, Idiopathic inflammatory myopathies, Quantitative sacroiliac scan, Whole-body bone scan, PM-Scl100t, Scintigraphic rehabilitation

Introduction

Sacroiliitis is an aching inflammation of the specific sacroiliac (SI) joint [1]. The inflammation may have different reasons, such as autoimmune disorders, microtrauma, malpractical exercise with periostitis [2-4], and infections like tuberculosis and brucellosis [5]. Sacroiliitis can also be related with Crohn's disease, ulcerative colitis, hyperuricemic gout, polychondritis, Whipple's disease, lumbar facet joint syndrome [6,7] and seronegative spondyloarthropathy like inflammatory bowel disease [8.9] or undifferentiated spondyloarthritis [10]. Clinical appearance of sacroiliitis is lower back pain (LBP) which may extend downward the legs, and many SI joint-associated disorders, for instance, postural impairment [11] and sciatica [12-14].

The idiopathic inflammatory myopathies (IIM) are a heterogeneous group of diseases, jointly appointed myositis, contributing symptoms of muscle weakness and inflammation in musculacture(s) [15]. Although there are a few case reports discussing autoimmune diseases or infections which might lead to myositis and sacroiliitis [9,16-18], there is currently no case report focusing on the relationship between myositis and sacroiliitis. Herein, we present a patient with overlapping syndromes of sacroiliitis and mild form of IIM as defined by clinical features.

Case Presentation

This is a 57-year-old male patient with underlying disease of dyslipidemia. He suffered from lower back and right knee pain for months. His knee pain often worsened after frequent squatting, which was inevitable as being a tour guide in Vietnam. Additionally, morning pain and stiffness was also stated by the patient. Besides, his neck pain had progressed over time. Right elbow pain extending to his forearm for two months was also complained by the patient.

Upon examination, he had local tenderness over his neck, upper back, right elbow, right forearm and bilateral SI joints. Full active assistive range of motion of right knee was tested. There was no crepitus or swelling over his legs. There was patellofemoral compression pain over his right knee. Hematological examination showed mild elevated HbA1c (6.2%). His electrolyte, white blood cell and platelet counts were all within normal limits. Besides, all rheumatoid examinations revealed negative findings, including ANA, P-ANCA, and HLA-B27. Anti-streptolysin O (ASLO) titer and CRP were both negative. Significantly, sixteen specific antibodies of myositis, e.g. Mi-2α, Mi-2β, TIF1γ, MDA5, NXP2, SAE1, Ku, PM-Scl100, PM-Scl75, Jo-1, SRP, PL-7, PL-12, EJ, OJ, Ro-52, were all negative, except PM-SCl 100 (Figure 1).

As for his image, the radiography of cervical spine showed degenerative change with disc space narrowing, marginal osteophyte formation and mild neuroforaminal stenosis between C3-C4, C4-C5, C5-C6, C6-C7. There was no radiographic evidence of bone lesion or fracture in the radiography of right elbow. Whole body bone scan, SPECT/CT, and quantitative SI scan (QSS) were also arranged for thorough survey. QSS was performed intravenously by injection of 750 MBq Tc-99m methylene diphosphonate, and counts of planar imaging of the sacrum/SI joints attain in the antero-posterior projections three hours later. There was elevated uptake at bilateral SI joints in both whole body bone scan and QSS (Figure 2 and 3). QSS, the SI joint-to-sacrum ratio (SI ratio), showed the inferior

third part of his SI joint was 1.91 in the right and 1.88 in the left. Aside from the findings mentioned above, no definite abnormal uptake elsewhere was noted.

Combined with his clinical symptoms and radiographic findings, this patient was diagnosed with ankylosing hyperostosis in cervicothoracicregion. Additionally, sacroiliitis was confirmed with QSS and bone scan, which could be correlated with his frequent LBP. With the positive finding of the sixteen antigens/antibodies myositis examination, he was also diagnosed with mild form of IIM. He was then under rehabilitation program, including physical therapy, shortwave diathermy, low power laser, and interferential therapy. Muscle relaxant and pain killer were also given for symptom relief.

Figure 1. Myositis 16 specific antibody testing. Only Sml-100 was positive, while the others were all negative.

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Antigen	.	Intensity	Class
Mi-2α	:	1	0
Mi-2β	:	1	0
TIF1γ	:	1	0
MDA5	:	0	0
NXP2	:	1	0
SAE1	:	0	0
Ku	:	1	0
PM-Scl100	:	6	(+)
PM-Scl75	:	0	0
Jo-1	:	0	0
SRP	:	1	0
PL-7	:	0	0
PL-12	:	1	0
EJ	:	0	0
OJ	:	0	0
Ro-52	:	0	0
Control	:	125	+++
Label(La)	:	-1	0

Figure 2. Whole body bone scan. Increased intake of bilateral sacroiliac joints is noticed.

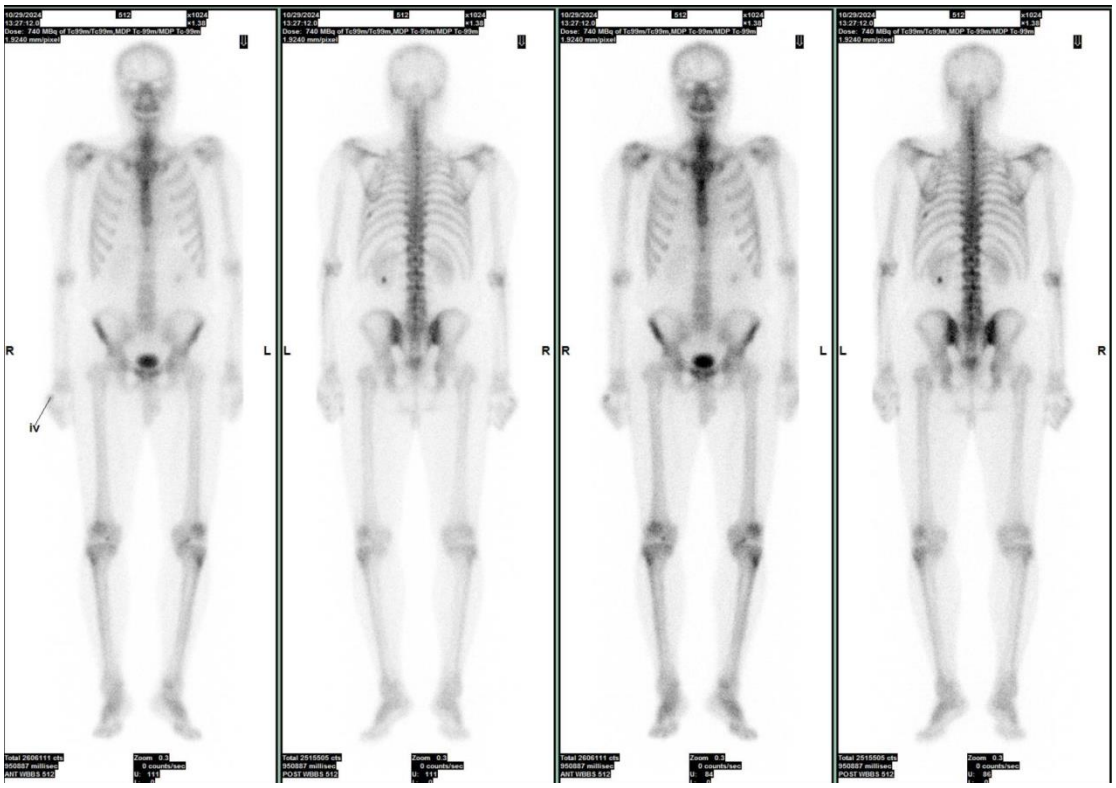
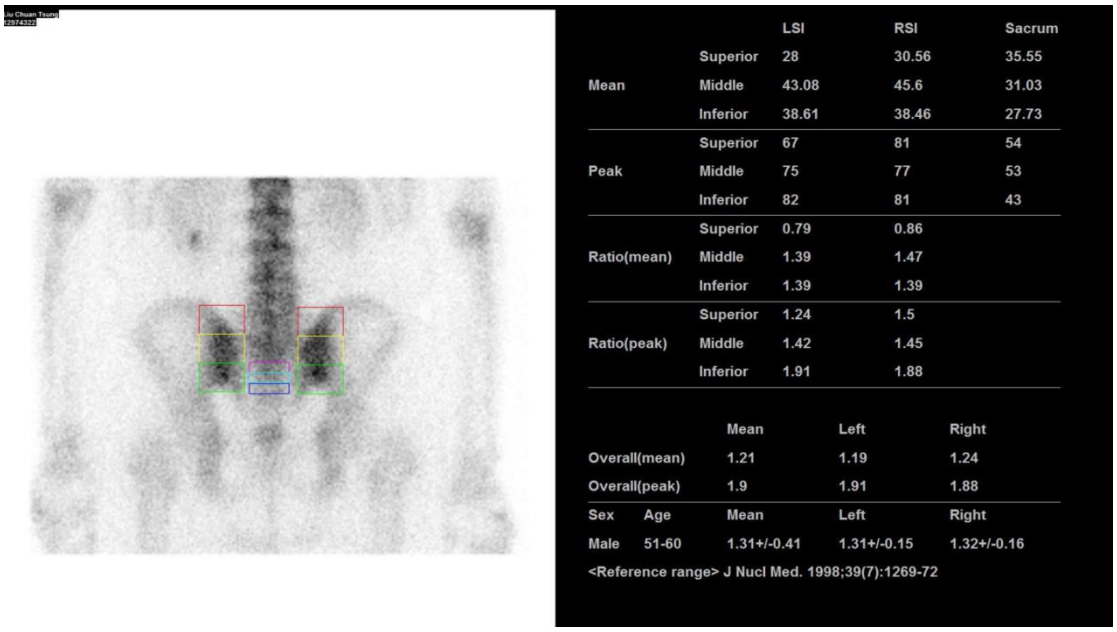


Figure 3. Quantitative sacroiliac scan. Elevated SI ratios of middle and inferior bilateral SI joints are detected.



		LSI	RSI	Sacrum
Mean	Superior	28	30.56	35.55
	Middle	43.08	45.6	31.03
	Inferior	38.61	38.46	27.73
Peak	Superior	67	81	54
	Middle	75	77	53
	Inferior	82	81	43
Ratio(mean)	Superior	0.79	0.86	
	Middle	1.39	1.47	
	Inferior	1.39	1.39	
Ratio(peak)	Superior	1.24	1.5	
	Middle	1.42	1.45	
	Inferior	1.91	1.88	

	Mean	Left	Right
Overall(mean)	1.21	1.19	1.24
Overall(peak)	1.9	1.91	1.88

Sex	Age	Mean	Left	Right
Male	51-60	1.31+/-0.41	1.31+/-0.15	1.32+/-0.16

<Reference range> J Nucl Med. 1998;39(7):1269-72

Discussion

This is a case of 57-year-old male patient who experienced LBP, and was then diagnosed as sacroiliitis by QSS and whole-body bone scan, and as mild form of IIM due to positive finding in his autoantibody testing of myositis. To our knowledge, this is the first case report that pointed out there is a correlation between sacroiliitis and mild form of IIM.

QSS together with bone scan has been reported as a essential indicator for inflammation of SI joint in the last six decades [19]. The SI/S ratio is calculated quantitatively by device computer according the region-of-interest method [20]. It was first reported by Buell et al. that increased SI ratios were subjected with SI illness/disorders [21]. In 1998, Kaçar et al. anounced the SI ratios as average of <1.32 for the healthy and <1.38 for patients as late arthritis, while up to 1.52–2.09 for patients as early arthritis [22]. Therefore, QSS with SI ratio can serve as an indicator for sacroiliitis [13,23] and has demonstrated in our studies. In our patient, the SI ratios of bilateral middle and inferior SI joints were all elevated. Combined with his clinical symptoms and image findings, sacroiliitis was then diagnosed.

Sacroiliitis occurs in varying situations, such as spondyloarthritis/spondyloarthropathy [10], osteitis condendsans ilii [12i], post-streptococcal reactive arthritis (PSRA) [24], and lower limb periostitis [2-4,13]. At first, axial spondyloarthritis (axSpA) was suspected for the reason that it was often presented with LBP [25]. According to the diagnostic criteria of axSpA by the assessment of SpondyloArthritis international Society, axSpA is considered while patients presenting with chronic LBP lasting for three months or more and age at onset of less than 45 years old [26,27]. However, the onset of his LBP was after his fifties. Additionally, it was found in the study of Poddubnyy et al. that nearly 90% of patients with spondyloarthritis is HLA-B27 positive [28]. Nevertheless, lab data of our patient showed negative finding of HLA-B27. With the negative result of his antibody test and clinical presentations, the diagnosis of axSpA would be less likely.

Upon reviewing other etiologies of sacroiliitis, PSRA was then suspected. Although there were case reports describing the axial involvement of PSRA, PSRA was more often confined with peripheral joints [29]. Clinically, it always instigates acute non-migratory polyarthritis with asymmetrical pattern, and tenosynovitis with peripheral arthritis in small joints may also take place [30]. It is also reported that there is a high correspondence between SI joint and elevated ASLO titers in PRSA patients [24,31,32]. Although the SI ratio of our patient was elevated, his ASLO titer was within normal limit. Also, PSRA presents a bimodal age distribution, that is, in the ages of 8-14 and 21-37 years [30]. Our patient was 57 years old, and the diagnosis of PRSA was excluded.

Testing for autoantibody is an essential tool for definite diagnosis of myositis. The anti-histidyl-transfer RNA synthetase (Jo1) autoantibodies, one of the myositis-specific autoantibodies ,was first added in the Tanimoto criteria for diagnosing IIM [33]. In 2015, myositis-associated antibodies (MSAs), e.g. anti-Ro52, anti-Ro60, anti-La, anti-U1RNP, PM-Scl and anti-Ku, were added in the new criteria of IIM by Troyanov et al. [34]. MSAs may acquaint with in some systemic autoimmune diseases, like in Sjögren´s syndrome, systemic sclerosis, and systemic lupus erythematosus [15].Observation on the serology examination of our patient, we could see positive result of PM-Scl only.

However, ANA, P-ANCA, and HLA-B27 were all negative. Additionally, the patient did not present with dry eyes, dry mouth or abnormal skin changes. As a result, the aforementioned diseases were less likely. Combined his muscle weakness with the result of autoantibody testing, mild form of IIM was then diagnosed.

Regarding the relationship of myositis and sacroiliitis, it is stated that myositis with sacroiliitis might be the result of different etiologies. There were studies indicating that malignancy might be associated with dermatomyositis and arthropathies, such as sacroiliitis [35,36]. Additionally, there was a case report introducing an unusual complication of brucellosis, which was piriformis myositis with sacroiliitis [17]. Although with low prevalence, it was also reported that dermatomyositis with symptomatic bilateral sacroiliitis can be caused by systemic lupus erythematosus [9]. However, there was no sign of infections, malignancy, or rheumatoid diseases in our patient. As a result, he was diagnosed sacroiliitis coinciding with mild form of IIM, without other underlying diseases mentioned above. Is it sacroiliitis a prodromal sign of this kind of myopathy? It is still far away to describe their relationship.

Limitation

This is a case report introducing the relationship between sacroiliitis and mild form of IIM. However, its limited sample size could not give answer to the prevalence of the conjuncture of these two diseases. Additionally, it is still unclear whether the observation of this case report would happen in other population out of Asia.

Conclusion

This is the first case report that pointed out there is a correlation between sacroiliitis and mild form of IIM, without cofounding factors of other rheumatoid diseases, malignancy, or infections. Future investigation is needed for more thorough literature review and greater sample size. Application of nuclear medicine testing is practical in the field of scintigraphic rehabilitation.

References

1. Lee, A., M. Gupta, K. Boyinepally, P.J. Stokey, and N.A. Ebraheim, Sacroiliitis: A Review on Anatomy, Diagnosis, and Treatment. Adv Orthop, 2022. 2022: p. 3283296.
2. Hui-Yi Su, Yu-An Hu, Wei-Chun Hsu, Shih-Chi Li, Shin-Tsu Chang. Effects of low power laser on pain and balance dysfunction caused by periostitis of lower limb: a case series. Taiwan Journal of Physical Medicine and Rehabilitation 2013 March; 41(1):51-57.
3. Cheng-Chiang Chang, Chih-Hung Ku, Chun-Sheng Hsu, Shin-Tsu Chang. Improvement of sacroiliac joint stress bottom-up after convalesce of foot periostitis: A randomized controlled trial. Asia Life Sciences 2016 January-July; 25(1):137-149.
4. Chen-Ling Chen, Hua-Ying Chuang, Cheng-Chiang Chang, Chih-Hung Ku, Shin-Tsu Chang. Randomized controlled trial of laser therapy versus conventional therapy in the treatment of medial tibial stress syndrome. International Journal of Orthopedics and Rehabilitation 2016 December; 3(2): 43-50.

5. Papadopoulos, E., P.J. Papagelopoulos, O.D. Savvidou, M.E. Falagas, G.S. Sapkas, and E.G. Fragkiadakis, Tuberculous sacroiliitis. Orthopedics, 2003. 26(6): p. 653-7; quiz 658-9.
6. Danielle Anne G. Tee, Shin-Tsu Chang. Appearance of facet joint syndrome in a case of chronic sacroiliitis: Evidenced from quantitative sacroiliac scintigraphy and dual images of SPECT-CT. International Journal of Frontiers in Biology and Pharmacy Research 2024, 05(01):019-023. DOI: https://doi.org/10.53294/ijfbpr.2024.5.1.0028.
7. Chun-Yi Lin, Ta-Chung Chao, Chuang-Hsin Chiu, Yu-Chiao Kuo, Shin-Tsu Chang. Coexistence of sacroiliitis and facet joint syndrome in an aged lady of Sjögren's syndrome: unexpected involvement of symphysis pubis. International Journal of Clinical Studies & Medical Case Reports 2024 October 30; 45(2):005. DOI:10.46998/IJCMCR.2024.45.001110.
8. Baronio, M., H. Sadia, S. Paolacci, D. Prestamburgo, D. Miotti, V.A. Guardamagna, G. Natalini, and M. Bertelli, Etiopathogenesis of sacroiliitis: implications for assessment and management. Korean J Pain, 2020. 33(4): p. 294-304.
9. Chandrasekhara, P.K., N.V. Jayachandran, J. Thomas, and G. Narsimulu, Systemic lupus erythematosus and dermatomyositis with symptomatic bilateral sacroiliitis: an unusual and interesting association. Mod Rheumatol, 2009. 19(1): p. 84-6.
10. Te-Jung Liu, Cheng-Chiang Chang, Liang-Cheng Chen, Heng-Yi Chu, Chun-Sheng Hsu, Shin-Tsu Chang. Relationship of HS CRP and sacroiliac joint inflammation in undifferentiated spondyloarthritis. Open Medicine 2018 May 19; 13(1):113-118.
11. Cheng-Chiang Chang, Heng-Yi Chu, Shang-Lin Chiang, Tsung-Ying Li, Shin-Tsu Chang Impairment of static upright posture in subjects with undifferentiated arthritis in sacroiliac joint in conjunction with elevation of streptococcal serology. Journal of Back and Musculoskeletal Rehabilitation 2009; 22(1):33-41.
12. Shin-Tsu Chang, Yuan-Tsan Wu, I-Hsuan Tsai, Liang-Cheng Chen, Heng-Yi Chu. Relief of sciatica after sacroiliac joint injection in a female with osteitis condensans ilii. Asia Life Sciences 2016 January-July; 25(1):511-515.
13. Chang, S.-T., The role of quantitative sacroiliac scintigraphy in clinical relevance: a literature review. MOJ Orthopedics & Rheumatology, 2023. 15(6): p. 233–237.
14. Yu-Chiao Kuo, Chuang-Hsin Chiu, Ta-Chung Chao, Chun-Yi Lin, Shin-Tsu Chang. Sciatica indirectly caused by sacroiliitis due to involvement of piriformis muscle: deduced from images of skeletal scintigraphy. International Journal of Research and Innovation in Applied Science (IJRIAS) 2024 November 14; IX (X): 271-276. DOI: https://doi.org/10.51584/IJRIAS.2024.910027.
15. Lundberg, I.E., F.W. Miller, A. Tjärnlund, and M. Bottai, Diagnosis and classification of idiopathic inflammatory myopathies. J Intern Med, 2016. 280(1): p. 39-51.
16. Van Tol, A.L., B. Tang, and I.D. Mackie, A case of Streptococcus canis bacteremia, osteomyelitis, sacroiliitis, myositis, and abscess. BMC Infect Dis, 2022. 22(1): p. 621.
17. Sandakly, N., G. El Koubayati, A. Ayoub, and F. Haddad, Brucellosis complicated by piriformis myositis and sacroiliitis: A case report and a review of the literature. Heliyon, 2024. 10(7): p. e28617.
18. Gómez Rodríguez, N. and O. Durán Muñoz, [Pyomyositis, sacroiliitis and spondylodiscitis caused by Staphylococcus hominis in a immunocompetent woman]. An Med Interna, 2006. 23(12): p. 582-4.
19. Verlooy, H., L. Mortelmans, S. Vleugels, and M. De Roo, Quantitative scintigraphy of the sacroiliac joints. Clin Imaging, 1992. 16(4): p. 230-3.

20. Davis, M.C., D.A. Turner, J.R. Charters, H.E. Golden, A. Ali, and E.W. Fordham, Quantitative sacroiliac scintigraphy. The effect of method of selection of region of interest. Clin Nucl Med, 1984. 9(6): p. 334-40.
21. Buell, U., The scintigraphic investigation of sacroiliac disease: influence of age on the uptake ratio between sacroiliac and os sacrum. J Nucl Med, 1977. 18(12): p. 1241-2.
22. Kaçar, G., C. Kaçar, B. Karayalçin, F. Güngör, T. Tuncer, and M. Erkiliç, Quantitative sacroiliac joint scintigraphy in normal subjects and patients with sacroiliitis. Ann Nucl Med, 1998. 12(3): p. 169-73.
23. Zhu Wei Lim, Shih-Chuan Tsai, Yi-Ching Lin, Yuan-Yang Cheng, Shin-Tsu Chang. A worthwhile measurement of early vigilance and therapeutic monitor in axial spondyloarthritis: a literature review of quantitative sacroiliac scintigraphy. European Medical Journal (EMJ) Rheumatology 2021 July 15; 8[1]:129-139.
24. Chang, S.T., C.H. Ku, and S.C. Cherng, Evidence-based correlation between anti-streptolysin O serum titer and sacroiliac joint disorder. J Rheumatol, 2007. 34(8): p. 1746-52.
25. Sieper, J. and D. Poddubnyy, Axial spondyloarthritis. Lancet, 2017. 390(10089): p. 73-84.
26. Rudwaleit, M., D. van der Heijde, R. Landewé, J. Listing, N. Akkoc, J. Brandt, J. Braun, C.T. Chou, E. Collantes-Estevez, M. Dougados, F. Huang, J. Gu, M.A. Khan, Y. Kirazli, W.P. Maksymowych, H. Mielants, I.J. Sørensen, S. Ozgocmen, E. Roussou, R. Valle-Oñate, U. Weber, J. Wei, and J. Sieper, The development of Assessment of SpondyloArthritis international Society classification criteria for axial spondyloarthritis (part II): validation and final selection. Ann Rheum Dis, 2009. 68(6): p. 777-83.
27. Rudwaleit, M., R. Landewé, D. van der Heijde, J. Listing, J. Brandt, J. Braun, R. Burgos-Vargas, E. Collantes-Estevez, J. Davis, B. Dijkmans, M. Dougados, P. Emery, I.E. van der Horst-Bruinsma, R. Inman, M.A. Khan, M. Leirisalo-Repo, S. van der Linden, W.P. Maksymowych, H. Mielants, I. Olivieri, R. Sturrock, K. de Vlam, and J. Sieper, The development of Assessment of SpondyloArthritis international Society classification criteria for axial spondyloarthritis (part I): classification of paper patients by expert opinion including uncertainty appraisal. Ann Rheum Dis, 2009. 68(6): p. 770-6.
28. Poddubnyy, D. and J. Sieper, Similarities and differences between nonradiographic and radiographic axial spondyloarthritis: a clinical, epidemiological and therapeutic assessment. Curr Opin Rheumatol, 2014. 26(4): p. 377-83.
29. Dagan, A., S. Dahan, A. Shemer, P. Langevitz, T. Hellou, T. Davidson, Y. Shoenfeld, and O. Shovman, Acute onset of psoriatic spondyloarthritis as a new manifestation of post-streptococcal reactive arthritis: a case series. Clin Rheumatol, 2019. 38(9): p. 2367-2372.
30. Bawazir, Y., T. Towheed, and T. Anastassiades, Post-Streptococcal Reactive Arthritis. Curr Rheumatol Rev, 2020. 16(1): p. 2-8.
31. Sarakbi, H.A., M. Hammoudeh, I. Kanjar, S. Al-Emadi, S. Mahdy, and A. Siam, Poststreptococcal reactive arthritis and the association with tendonitis, tenosynovitis, and enthesitis. J Clin Rheumatol, 2010. 16(1): p. 3-6.
32. Uziel, Y., L. Perl, J. Barash, and P.J. Hashkes, Post-streptococcal reactive arthritis in children: a distinct entity from acute rheumatic fever. Pediatr Rheumatol Online J, 2011. 9(1): p. 32.
33. Tanimoto, K., K. Nakano, S. Kano, S. Mori, H. Ueki, H. Nishitani, T. Sato, T. Kiuchi, and Y. Ohashi, Classification criteria for polymyositis and dermatomyositis. J Rheumatol, 1995. 22(4): p. 668-74.
34. Troyanov, Y., I.N. Targoff, J.L. Tremblay, J.R. Goulet, Y. Raymond, and J.L. Senécal, Novel classification of idiopathic inflammatory myopathies based on overlap syndrome features and autoantibodies: analysis of 100 French Canadian patients. Medicine (Baltimore), 2005. 84(4): p. 231-249.

35. Mody, G.M. and B. Cassim, Rheumatologic manifestations of malignancy. Curr Opin Rheumatol, 1997. 9(1): p. 75-9.
36. Naschitz, J.E., I. Rosner, M. Rozenbaum, E. Zuckerman, and D. Yeshurun, Rheumatic syndromes: clues to occult neoplasia. Semin Arthritis Rheum, 1999. 29(1): p. 43-55.

Citation: Chia-Ling Tsao, Daniel Hueng-Yuan Shen, Wen-Hsuan Hsiao, Shin-Tsu Chang, Adv Clin Med Sci, "Is it Coincidence of Sacroiliitis and a Mild Form of Idiopathic Inflammatory Myopathies? A Case Report". 2024; 2(2): 112

Received Date: December 09, 2024; Published Date: December 13, 2024