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Case Report

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Sacroiliitis attributed to post-streptococcal reactive arthritis and congenital absence of the fifth lumbar vertebra: diagnosis and scintigraphic findings

Abstract

Post-streptococcal reactive arthritis (PSRA) is associated with a prior group A streptococcal infection and has a bimodal age distribution. Clinical features of arthritis caused by PSRA, including symmetric or asymmetric, usually non-migratory, can affect any joint, persistent or recurrent, and the sacroiliac joint is most affected in Asia. Congenital lumbosacral malformation including transitional vertebrae or spina bifida occulta, is one of the etiologies of lower back pain. However, the congenital absence of the fifth lumbar vertebra with lower back pain has not been reported yet. We present a case of a 28-year-old man having only four lumbar vertebrae, who was ultimately diagnosed with PSRA involving the SI joints. We assumed that his lower back pain is not solely due to PSRA, the absence of the fifth lumbar vertebra also plays an important role. A detailed description of these two mechanisms which may contribute to his symptom was discussed in the text.

Keywords: sacroiliitis, post-streptococcal reactive arthritis, anti-streptolysin o, quantitative sacroiliac scintigraphy, whole body bone scan, single photon emission computed tomography – computed tomography, scintigraphic rehabilitation

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Introduction

Post-streptococcal reactive arthritis (PSRA) has been defined as more than 1 joints, associated with a prior group A streptococcal infection in a patient who does not meet the Jones criteria for the diagnosis of acute rheumatic fever.[1] It has a bimodal age allocation by ages 8-14 and 21-37 years with equal ratio in male and female[2] and can take place within 10 days of group A streptococcal infection.[3] A previous report summarized that the clinical features of arthritis caused by PSRA, including acute onset with non-migratory, symmetric or asymmetric, can affect any appendicular joint, persistent or recurrent, and the involved arthritis is poorly work with salicylates or nonsteroidal anti-inflammatory drugs.[4] Corticosteroid is the first-line treatment option for PSRA.[2] While most patients resolved spontaneously within some weeks, part of cases may persist longer or recur over time.[2] PSRA has been demonstrated with frequent involvement of the sacroiliac (SI) joints in Asia.[5]

Congenital lumbosacral malformations associated with lower back pain (LBP) lasting for more than 4 weeks including transitional vertebrae and SBO, accounted for one-tenth of the patients with LBP,[6] highlighting the significant role of congenital lumbosacral malformations in the etiology of LBP. However, the congenital absence of the fifth lumbar vertebra in a patient with LBP has not been reported yet.

We herein report a case of a young man with PSRA with involvement of SI joints, who lacks the fifth lumbar vertebra. According to the skeletal scintigraphic findings, he had increased uptake over the bilateral SI joints, which might be related to his LBP.

Case presentation

A 28-year-old man visited Outpatient Department of Rehabilitation of our hospital for his LBP on September 14th, 2024. The patient said the pain has lasted for one more year, and it would exaggerate after running. He also mentioned that the situation has worsened over the past week. Physical examination revealed local tenderness of lower back and SI joints. At OPD, X-ray films of lumbar spine showed suspected absence of the fifth lumbar vertebra with no visible lesion (Figure 1).

To gather more detailed information about his illness, we scheduled a series of tests, including Whole Body Bone Scan (WBBS), Quantitative Sacroiliac Scintigraphy (QSS), Single Photon Emission Computed Tomography-Computed Tomography (SPECT-CT) of bone, and X-ray films of thoracic spine. WBBS and SPECT-CT revealed an increased uptake of Tc -99m methylene diphosphonate (Tc-99mMDP) in the bilateral SI joints (Figure 2 & 3). QSS showed a left SI/S ratio of 1.7 and a right SI/S ratio of 1.64.[7,8] The X-ray films of thoracic spine presented that there are twelve pairs of ribs (Figure 4), confirming that this patient has only four lumbar vertebrae rather than the typical five, with the thirteenth pair of ribs attaching to the first lumbar vertebra. The results of the following tests of serum sample were all negative: white blood cell count, erythrocyte sedimentation rate, high-sensitive C-reactive protein, antinuclear antibody, rheumatoid factor, and human leukocyte antigen-B27. However, anti-streptolysin O (ASLO) titer was measured at 347 IU/mL, exceeding the normal upper limit of 116 IU/mL.

Figure 1 The X-ray films of lumbar spine showed suspected absence of the fifth lumbar vertebra with no visible lesion.

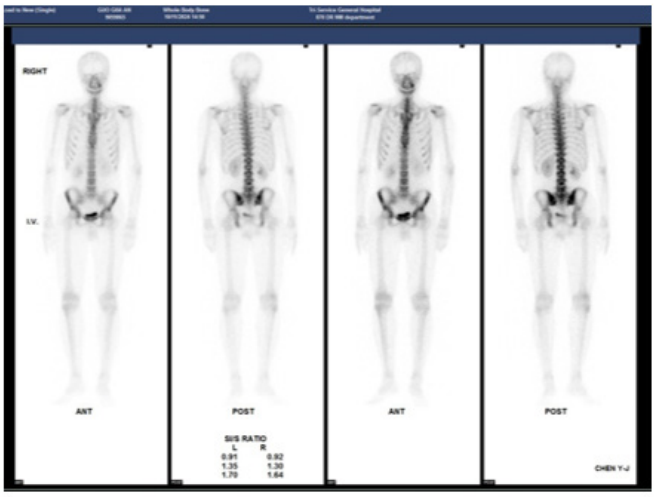

Figure 2 The WBBS of the patient could see an increased uptake over the bilateral SI joints, suggesting inflammatory changes.

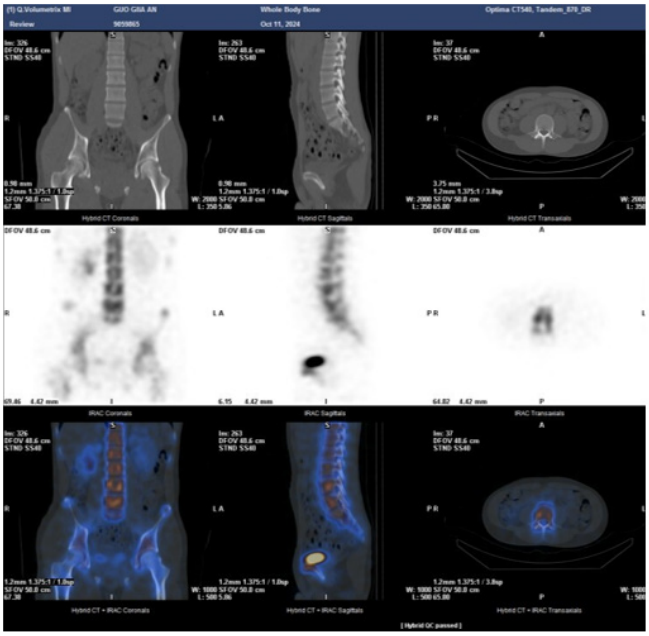

Figure 3 The top row shows CT scans, the middle row shows SPECT scans, and the bottom row displays fused SPECT-CT images. From left to right, the images are organized into coronal, sagittal, and axial views. The enhancements of the hybrid SPECT-CT at the bilateral SI joints, in favor of PSRA with involvement of SI joints

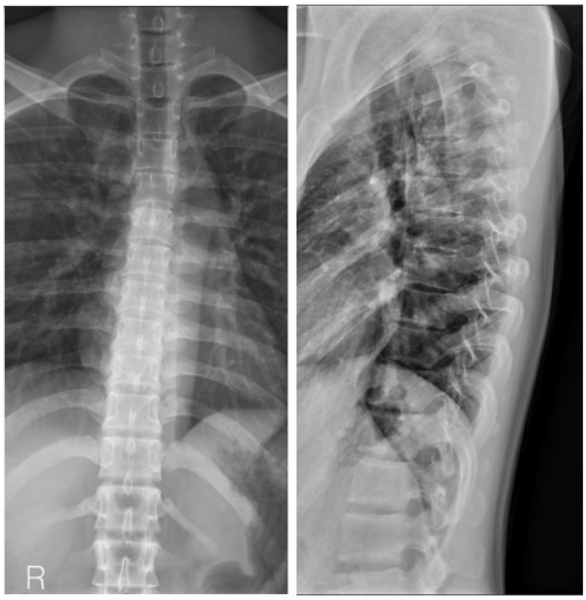

Figure 4 The X-ray films of thoracic spine showed that there are 12 pairs of ribs.

After being diagnosed PSRA with bilateral sacroiliitis along with congenital absence of the fifth lumbar vertebra on October 26th, 2024, the patient received NSAIDs, muscle relaxants, and non-pharmacological interventions, such as wearing a military combat belt (S belt in Taiwanese army), maintaining proper posture, avoiding excessive weight-bearing, and engaging in back muscle training. In subsequent outpatient follow-up visits, he reported an improvement in his LBP.

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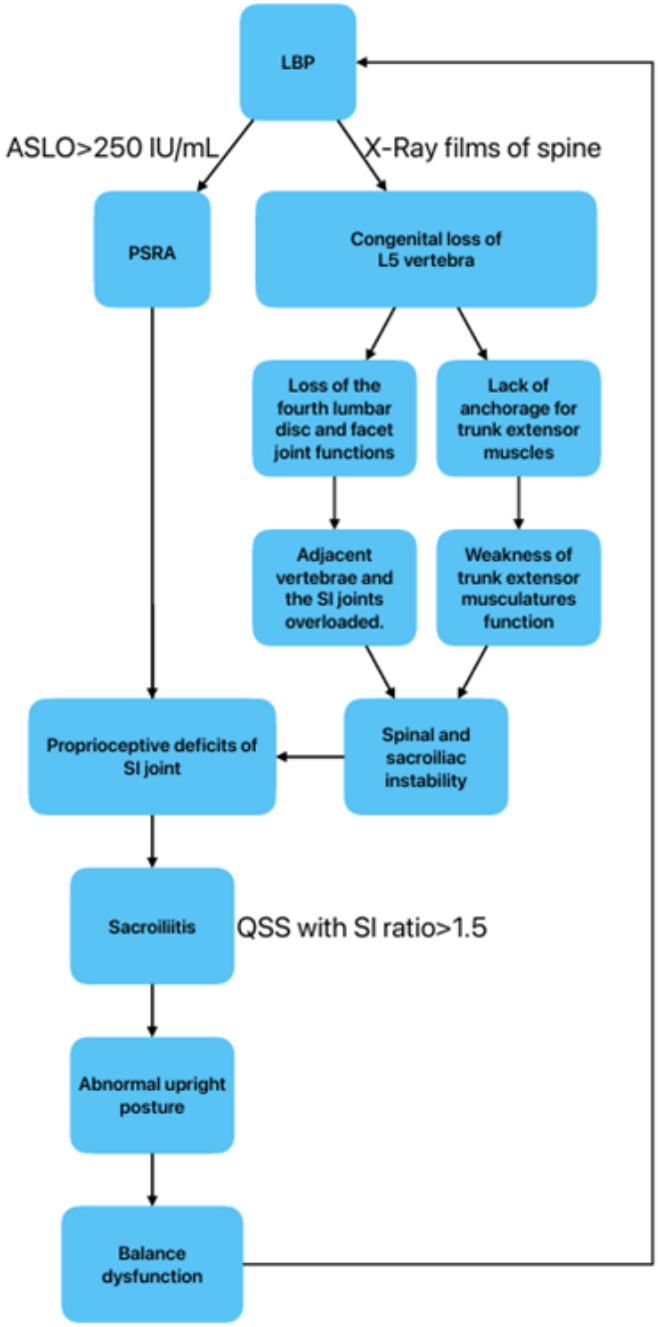

Figure 5 Schematic illustration of the pathophysiology of post-streptococcal reactive arthritis, congenital loss of L5 vertebra, and their related disorders.

LBP: lower back pain; ASLO: antistreptolysin O; PSRA: post-streptococcal reactive arthritis; SI: sacroiliac; QSS: quantitative sacroiliac scintigraphy

Discussion

In this case, the patient presented with elevated ASLO titers, indicating a prior group A streptococcal infection, and was diagnosed with PSRA with involvement of the SI joints. The elevated ASLO titers in combination with PSRA affecting the SI joints suggest that this inflammatory mechanism may be contributing to his chronic LBP, alongside the unusual spinal structure. The congenital absence of the fifth lumbar vertebra (L5), resulting in only four lumbar vertebrae (L1-L4), is an unusual anatomical variation rarely reported in medical literature, and can be one reason contributing LBP. To our knowledge, this is the first report to show a patient with sacroiliitis due to both PSRA and four lumbar vertebrae (L1-L4) only.

Initial X-ray films of lumbar spine in this case showed the presence of only four lumbar vertebrae, with no transitional vertebra noted. Given that the vertebra with rib attachments is classified as thoracic vertebra, and the remaining vertebrae without rib attachments number only four, it was initially presumed that this patient had only four lumbar vertebrae. Subsequent X-ray films of thoracic spine were conducted to confirm the presence of twelve pairs of ribs, excluding the possibility of the thirteenth pair of ribs attaching to the first lumbar vertebra. These findings verified that this patient indeed has only four lumbar vertebrae and 12 pairs of ribs, without any transitional vertebrae.

The increased ASLO titer indicates a past infection with group A streptococcus, even subclinical or asymptomatic,[9,10] which titer is valuable markers for early identification of PSRA.[11,12] PSRA can affect typically involved varying appendicular and axial joints.[13] In Taiwan, researchers have shown that SI joint involvement can be part of the disease progression in PSRA, which study found a very strong link between ASLO titers and SI/S ratios,[5,14] indicating that SI joint involvement is a manifestation of PSRA. Although our patient might not have experienced obvious symptoms of the initial infection, the immunological response leading to PSRA and ensuing sacroiliitis likely contributed to the current presentation of LBP.

Sacroiliitis, or inflammation of the SI joints, is a well-recognized essence of LBP,[15] which can be caused by varying reasons,[16–20] and is highly related to other disorders.[21,22] In this case, the increased uptake of Tc-99m MDP in both SI joints observed on WBBS and SPECT-CT images indicated that there might be inflammation on the patient's bilateral SI joints. Considering the patient's elevated ASLO titer, this finding supports that sacroiliitis here is a manifestation of PSRA, further substantiated by SI/S ratios of QSS, which reflect inflammation intensity. The chronicity and exacerbation of pain post-running also align with sacroiliitis, as mechanical stress on inflamed SI joints can worsen symptoms.[23] Furthermore, the use of NSAIDs has been shown to help manage sacroiliitis symptoms by reducing inflammation,[2] providing symptomatic relief in our case.

The patient's LBP appears to be multifactorial, with two primary mechanisms contributing to his symptoms. The first mechanism is the inflammatory process of PSRA affecting the SI joints, resulting in sacroiliitis and associated pain.

The second mechanism is the congenital absence of the fifth lumbar vertebra, which reduces stability and alters the biomechanical distribution of forces in the lumbar spine. In this patient, the absent vertebra reduces the spine's ability to manage various forces - such as compression, tensile, shear, bending moments, and torsional moments - that typically occur during the movements of spine.[24,25] Under normal conditions, the lumbar spine and intervertebral discs form an effective load-bearing system that can adapt to external stresses.[26] The intervertebral discs consist of the inner nucleus pulposus facing hydrostatic pressure while loaded, and the outer annulus fibrosus providing outside resistance against the radial bulging of the nucleus pulposus stemmed from hydrostatic pressure, which acts as a shock absorber, and the inner and outer parts ensure flexibility to withstand mechanical loading, which can transfer loads and dissipate energy imposed on the spine, slowing down the transfer of forces and reducing stress on the vertebrae above and below maintain a baseline pressure that supports resistance to these forces.[26,27] The Intervertebral discs therefore play a key role in spinal mechanics, functioning as

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cushioning structures that help distribute forces and absorb impact between vertebrae during movement.[28] Facet joints also play a significant role in providing stability to the spine and they have been associated with LBP symptoms and other spinal disorders.[29] Yang & King[30] estimated that 3%-25% of the axial compressive load is transferred through the normal facet joints, based on the study using cadaveric lumbar specimens. The facet joints perform different biomechanical functions depending on the spinal position, including extension, forward flexion, axial rotation, and lateral bending.[31] In this case, the lack of the lumbar disc and the fifth lumbar vertebra interrupts the distribution of forces, potentially overloading adjacent vertebrae and contributing to increased strain on the SI joints. Also, the absence of a vertebral body at the typical L5 level is functionally like the instability seen in patients with spina bifida occulta, where muscles like the lower parts of multifidus and erector spinae lack a proper attachment site.[32] The lumbar back muscles are essential components of the spine's stabilizing system and play a significant role for segmental stability.[33,34] This lack of anchorage may impair control of postural sway owing to weakness of trunk extensor musculatures function like the patient of SBO,[35] further lead to spinal instability and increased susceptibility to LBP. Addressing both two mechanisms is essential in managing his condition, as each mechanism requires targeted interventions and ongoing monitoring. For PSRA, the main treatment is NSAIDs.[2] For the four lumbar vertebrae (L1-L4 only), our management includes regular exercise to strengthen the back muscles, protective devices such as lumbar belts, maintaining good postures, avoiding excessive weight loading, and regular follow-ups.[23]

Conclusion

In our case, both PSRA with involvement of SI joints and absence of the fifth lumbar vertebra are potential causes of LBP, necessitating specific management and follow-up for each individual cause. A careful and precise diagnosis is crucial in managing cases with rare pathological structure like this patient.

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None.

Conflicts of interest

The authors declare that there are no conflicts of interest.

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