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Case Report

Spontaneous Laminar Fracture during Successful Conservative Treatment of Lumbar Spondylolysis at the Adjacent Spinal Level: A Case Report

Hiroaki Manabe,[1] Toshinori Sakai,[1] Kosuke Sugiura,[1] Yoshihiro Ishihama,[1] Fumitake Tezuka,[1] Kazuta Yamashita,[1] Yoichiro Takata,[1] Toru Maeda,[1] and Koichi Sairyo[1]

Although lumbar spondylolysis (LS) is regarded as a stress fracture, the underlying pathomechanism has yet to be fully elucidated. Here, we present a case that casts doubt on the notion that LS is truly a stress fracture. An 11-year-old female basketball player was brought to our clinic with a 2-week history of persistent low back pain. Magnetic resonance imaging with short TI inversion recovery (STIR-MRI) showed high signal intensity changes at the L4 pedicles bilaterally. Computed tomography (CT) revealed a faint fracture line at the left pars interarticularis. We made a diagnosis of stress fracture and recommended conservative treatment, including cessation of sports activities and wearing of a hard brace. Compliance with treatment was excellent. As expected, the STIR-MRI findings at L4 gradually resolved and bone healing was achieved. However, a follow-up STIR-MRI scan 10 weeks later revealed high signal intensity at the left L5 pedicle. Conservative treatment was continued for the findings at L5, which were considered to indicate a stress fracture (spondylolysis). Five weeks later, CT revealed a bony defect in the lamina at L5 on the left and bone union at L4. Although LS is generally considered to be a stress fracture, there have been several reports of familial occurrence and genetic predisposition. This patient's mother had also been treated for spondylolysis at L5. These observations suggest an underlying genetic etiology in this case.

Keywords: low back pain, lumbar spondylolysis, laminar fracture, adolescents

Introduction

Lumbar spondylolysis (LS) has a reported incidence of approximately 6% in the general population.[1)] Biomechanical and clinical investigations indicate that LS is a pseudarthrosis that occurs as a result of a stress fracture[2)]; however, the pathophysiological mechanism involved has yet to be completely elucidated. Interestingly, in the early literature, LS was considered a hereditary disease rather than the result of a simple stress fracture.[3,4)] Presently, some recent literature suggest genetic factors as a cause of LS.[5)] In this report, we describe a patient who developed a spontaneous laminar fracture at L5 during successful conservative treatment for LS at L4. This case raises the question of whether or not spondylolysis is truly a stress fracture.

Case Report

An otherwise healthy 11-year-old girl who was a basketball team member visited our outpatient clinic with a 2-week history of persistent low back pain that was triggered by lumbar extension and right rotation. There was tenderness at the L4 spinous process and paravertebral muscles bilaterally. The Kemp test was positive on both sides. Magnetic resonance imaging with short TI inversion recovery (STIR-MRI) showed high signal intensity changes at the L4 pedicles bilaterally (Fig. 1A), and computed tomography (CT) revealed a faint fracture line at the left pars interarticularis (Fig. 1B). The diagnosis was stress fracture of the L4 lamina (very early-stage LS) on the right and early-stage LS on the left.[6)] Conservative treatment was recommended, including rest, cessation of sports activities, and wearing of a hard brace, specifically, a molded plastic thoracolumbosacral orthosis.[7,8)]

Compliance with conservative treatment was excellent; consistent with the healing process, the STIR-MRI findings at L4 gradually diminished and bone healing was achieved (Fig. 2A). However, despite the successful conservative treatment for the stress fracture of the L4 lamina, follow-up STIR-MRI scan 10 weeks later revealed high signal intensity at the left L5 pedicle, indicating that the LS originated from a stress fracture (Fig. 2B). Conservative treatment was continued for the new findings identified at L5. Five weeks later, CT revealed a complete ossification of the bony defect in the lamina at L5 on the left and bone union at L4 (Fig. 3).

Discussion

LS has been considered to be a stress/fatigue fracture of the pars interarticularis since the report by Wiltse et al. in 1975.[2)] An abundance of literature has been published in support of this theory, and LS is now thought to be a sports-related disorder.[9)] Clinical and biomechanical investigations have demonstrated that repetitive extension/rotation of the lumbar spine concentrates stress on the pars interarticularis and that these movements are likely to be risk factors for the development of LS outbreak.[10,11)] However, there are several studies suggesting a contribution of genetic factors which

[1]Department of Orthopedics, Institute of Biomedical Sciences, Tokushima University Graduate School, Tokushima, Tokushima, Japan

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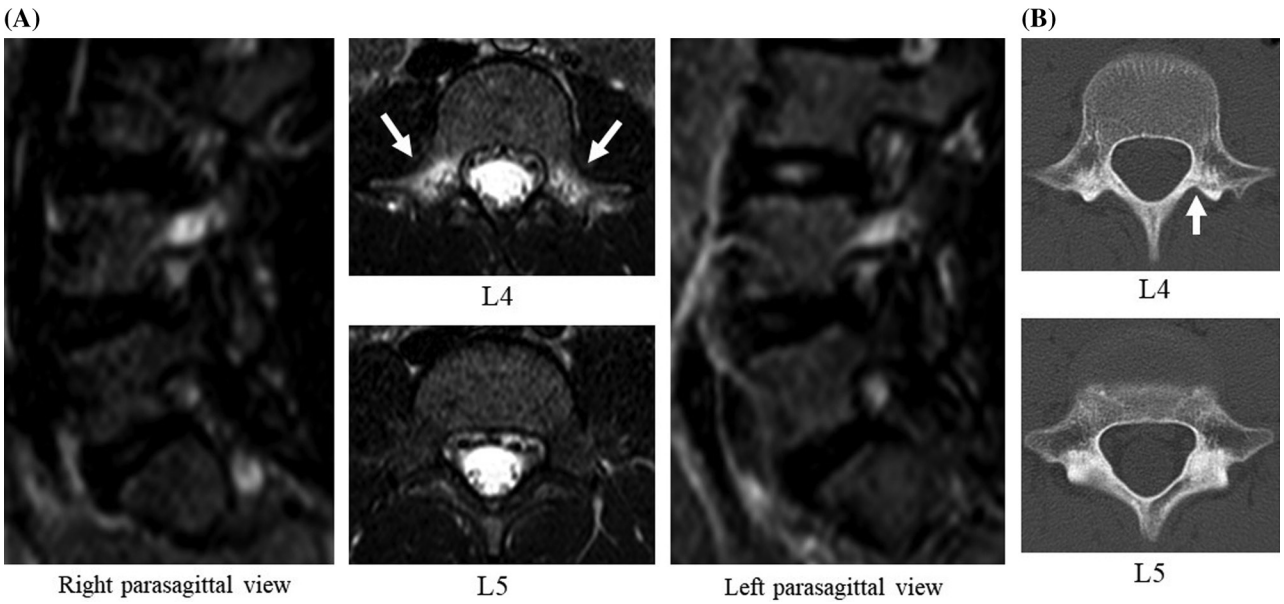

Fig. 1 (**A**) Magnetic resonance images showing a high signal intensity change at the L4 pedicles bilaterally (indicated by arrows). (**B**) CT scans showing a faint fracture line (arrow) at the L4 pars on the left with no findings at L5. CT: computed tomography.

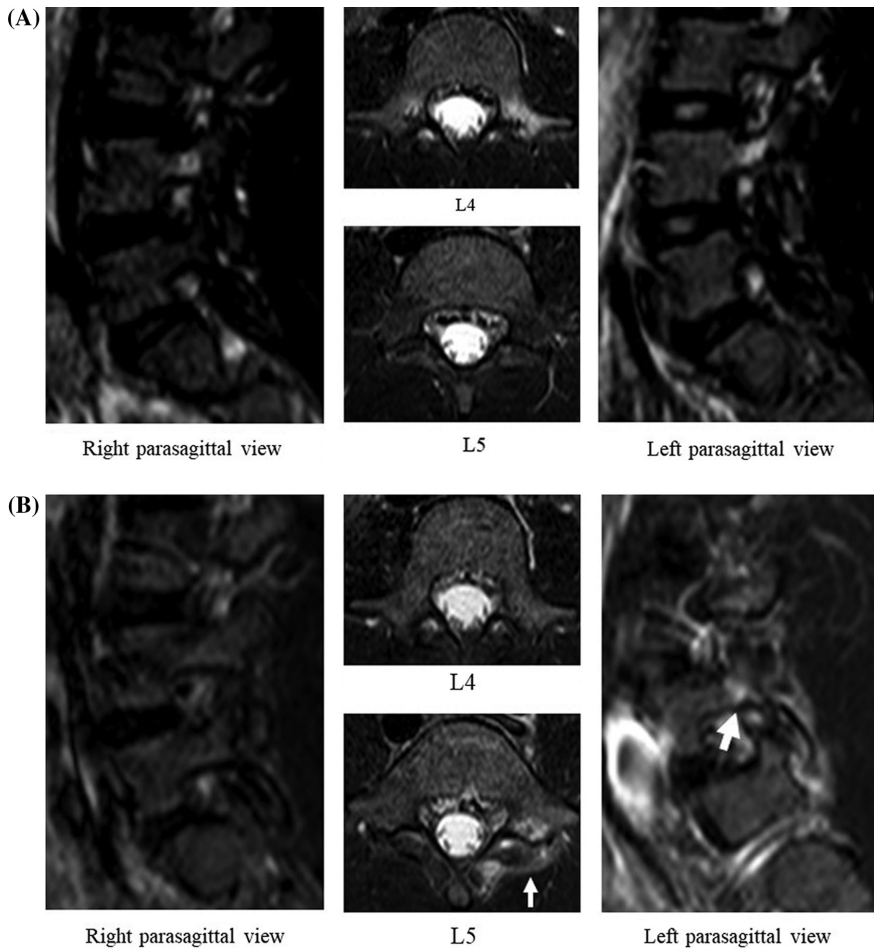

Fig. 2 (**A**) Magnetic resonance images obtained at follow-up 5 weeks later showing a decrease in the high intensity changes at the L4 pedicles bilaterally. (**B**) Magnetic resonance scans acquired 10 weeks later showing a new high intensity change at the L5 pedicle on the left. The arrows indicate extraosseous and bone marrow edema.

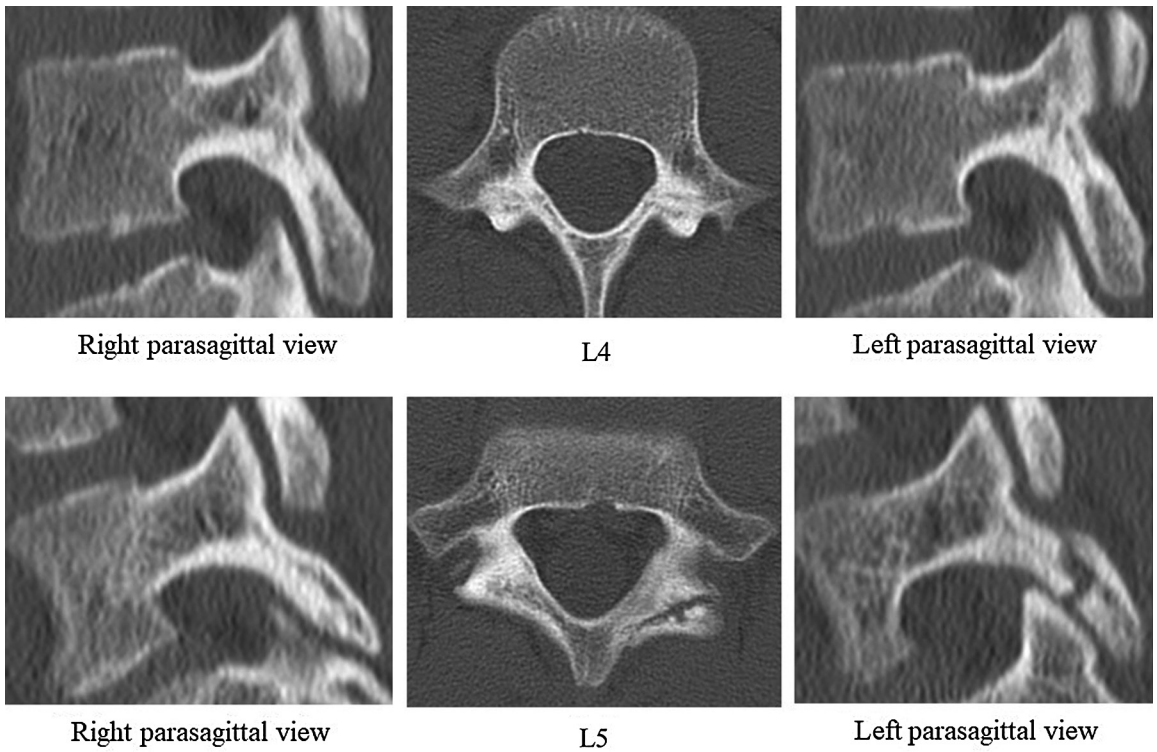

Fig. 3 CT scans obtained 15 weeks later showing bone union at L4 and complete ossification of the bony defect in the lamina at L5 on the left. CT: computed tomography.

show different prevalence by races and familial accumulation.[3,9,12] The prevalence of spondylolysis varies according to race and sex, with reported incidence rates of 6.4% for white men, 2.8% for black men, 2.3% for white women, and 1.1% for black women.[9] A particularly high incidence (of approximately 40%) has been identified in the Inuit population.[3,12] Several investigators have described a familial accumulation of LS.[4,13] Wynne-Davies and Scott investigated 147 first-degree relatives of 47 patients with spondylolysis and found that 19% had spondylolysis, which is markedly higher than the incidence in the general population.[13] These reports strongly suggest the role of a genetic predisposition in the pathogenesis of LS. Cai et al.[14] recently identified a novel heterozygous mutation in the sulfate transporter gene SLC26A2 in five affected subjects of a Chinese family. Furthermore, they reported that mutations in SLC26A2 were identified to cause a family of several recessive chondrodysplasias; however, the clinical abnormalities in their cases are restricted to cartilage and bone in the lumbosacral spine. Basically, their results provided an evidence that SLC26A2 specifically plays a role in the development of cartilage in lumbosacral spine.

In the present case, the lamina fracture (spondylolysis) at L5 was considered to have occurred spontaneously rather than as a result of minor trauma or playing sports, which was supported by the fact that the stress fracture of the lamina (spondylolysis) at L4 was successfully treated. Single-photon emission computed tomography (SPECT) was not taken for this case,[15,16] but it might provide some information for the etiology of this patient. Interestingly, the patient's mother, who was a volleyball player, had also been treated for L5 spondylolysis in her growth period. As it happens, she had consulted our clinic for her chronic low back pain and a diagnosis of spondylotic spondylolisthesis was made before this patient came to us (Fig. 4).

These observations suggest an underlying genetic etiology in this case.

We speculated that several factors other than genetic predisposition may have contributed to the LS at L5 in our patient. First, wearing of a hard brace could not fully protect the patient against the mechanical load generated by activities of daily living, such as running or cycling. Goto et al.[17] recently reported that sprinting increases the hip extension angle, spine rotation angle, and hip flexion moment, and can be a risk factor for LS. Furthermore, our patient was a Japanese elementary school student, and so the load generated by carrying the traditional heavy Japanese school backpack (*randoseru*) every day might have caused mechanical stress on the lumbar spine.[18] Second, the anatomic differences between L5 and the other spinal levels should be considered, in particular the blood supply. Tezuka et al.[19] identified four main anatomic variations in the blood supply to the posterior element (lamina) of L5, with a segmental artery being detected at this level on CT angiography in fewer than 10% of patients.

Further biomechanical, biological, genetic, and anatomic investigations are needed to clarify the pathogenesis of LS.

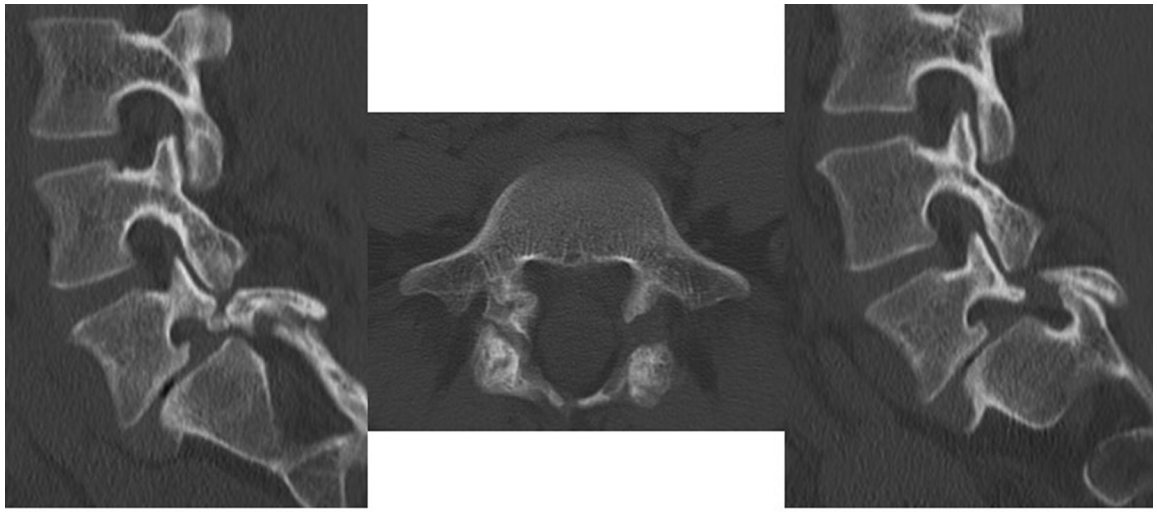

Fig. 4 CT scans showing terminal-stage spondylolysis at L5 in the patient's mother. CT: computed tomography.

Conflicts of Interest Disclosure

All authors report no conflicts of interest concerning this article.

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Corresponding author:
Toshinori Sakai, MD, PhD, Department of Orthopedics, Institute of Biomedical Sciences, Tokushima University Graduate School, 3-18-15 Kuramoto-cho, Tokushima, Tokushima 770-8503, Japan.
✉norinoridowluck@yahoo.co.jp