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ORIGINAL ARTICLE • SPINE - SPONDYLOLYSIS

Clinical features of patients with pars defects identified in adulthood

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Abstract
Purpose Lumbar spondylolysis is considered a stress fracture of the *pars interarticularis* that occurs during growth. However, it is sometimes insidious and identified in adults as pseudoarthrosis, the terminal-stage of spondylolysis. The purpose of this study was to identify the clinical features of patients with terminal-stage spondylolysis that first manifested during adulthood.
Patients and methods Thirty-six patients (21 men, 15 women; mean age 55.8 years; age range 25–77 years) with low back pain (LBP) were studied. In all patients, lumbar spondylolysis had not been diagnosed until the first visit to our hospital. Patient data collected were history of athletic activity and LBP during their growth period and radiological findings, such as spinal level, displacement, and spina bifida occulta (SBO).
Results Among the 36 patients, including a patient with multi-level spondylolysis (L4 and L5), a total of 37 vertebrae with terminal-stage spondylolysis were identified. Twenty-three (89.2 %) of the 37 vertebrae had L5 spondylolysis. Sixteen patients (44.4 %) had no history of athletic activity, 26 (72.2 %) had no experience of LBP during their growth period, and 14 (38.9 %) had neither. Twenty of the 37 vertebrae (70.4 %) involved displacement (grade 1 = 14; grade 2 = 6). In nine patients (25.0 %; eight men, one woman), SBO of the sacrum was accompanied by L5 spondylolysis.
Conclusions Approximately 90 % of patients with terminal-stage spondylolysis that was first diagnosed in adulthood involved the L5. Also, about 40 % had no history of athletic activity or experience of LBP during their growth period. In addition, only some patients with L5 spondylolysis had SBO, and all but one of these patients was male. This suggests that male patients with L5 spondylolysis may have some congenital predisposition.

Keywords Lumbar spondylolysis · Adult · *Pars interarticularis* · Spina bifida occulta · Pathophysiology

Introduction

Lumbar spondylolysis is considered a stress fracture of the *pars interarticularis* of the lumbar spine, as suggested by Wiltse et al. [1, 2]. Biomechanically and clinically, repetitive lumbar extensions and rotations that are required for athletic activity were regarded as the main risk factor [3, 4]. Most of the time, the stress fracture occurs in the adolescent period and is a frequent cause of low back pain (LBP) in teenagers and young adults, especially those who are involved in sports [5–7]. However, the onset is sometimes insidious, and the disorder is not identified until patients experience LBP in adulthood, when the spondylolysis presents as a pseudoarthrosis or terminal-stage spondylolysis.

A congenital predisposition to spondylolysis has also been suggested [7–10]. As a phenotype associated with spondylolysis, spina bifida occulta (SBO) had been reported to be a strongly associated disorder, particularly in a cohort of elementary school age children with L5 spondylolysis [9]. Recently, genetic analysis on the pathophysiology of dysplastic spondylolysis has been

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published [11]. However, the pathophysiology of spondylolysis has not been clearly elucidated. To address this question, we analyzed the clinical features of patients with terminal-stage spondylolysis that was first diagnosed in adulthood.

Patients and methods

Thirty-six consecutive patients who consulted our clinic for LBP between January 2012 and October 2014 were enrolled in this study. Patients <20 years old were excluded. In all patients, lumbar spondylolysis had not been diagnosed until the first visit to our hospital. Patient data were collected on sex difference, history of athletic activity and experience of LBP during their growth period, and radiological findings, such as spinal level, grade of displacement according to Meyerding system [12], and relationship between spondylolysis and SBO. An institutional review board exemption was obtained for this study.

Results

Spinal levels, stage of lumbar spondylolysis, and sex differences

Among 36 patients (mean age 55.8 years, age range 25–77 years), a total of 37 vertebrae with terminal-stage (pseudoarthrosis stage) spondylolysis were identified. All patients had bilateral spondylolysis, and there was one multi-level spondylolysis (L4 and L5). There was a slight male predominance (58.3 %), with a male–female ratio of 1.4:1 (Fig. 1). Spondylolysis was at the level of L5 in 33 of 37 vertebrae (89.2 %; 20 men, 13 women), L4 in two female patients, and L3 in two patients (one male, one female).

History of athletic activity and low back pain in childhood and adolescent period

Sixteen patients (44.4 %) had no history of athletic activity, while 26 (72.2 %) had no experience of LBP in childhood and adolescent period (Fig. 2). Fourteen patients (38.9 %) had a history of neither athletic activity nor LBP.

Isthmic (spondylolytic) spondylolisthesis

Twenty of the 37 vertebrae (54.1 %) involved displacement (grade I = 14, grade II = 6) (Fig. 3). There were 33 vertebrae with L5 spondylolysis, 17 (51.2 %) of which had displacement (grade I = 13, grade II = 4). Among the other vertebrae with L3 and L4 involvement, 3 (75.0 %) had displacement (grade I = 1, grade II = 2).

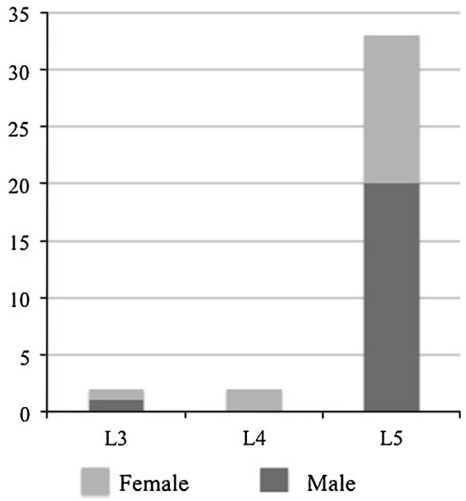

Fig. 1 Distribution of spondylolysis, based on affected lumbar vertebra and sex. Almost all spondylolyses involved the L5 vertebra. There was a slight male predominance (58.3 %), with a male–female ratio of 1.4:1

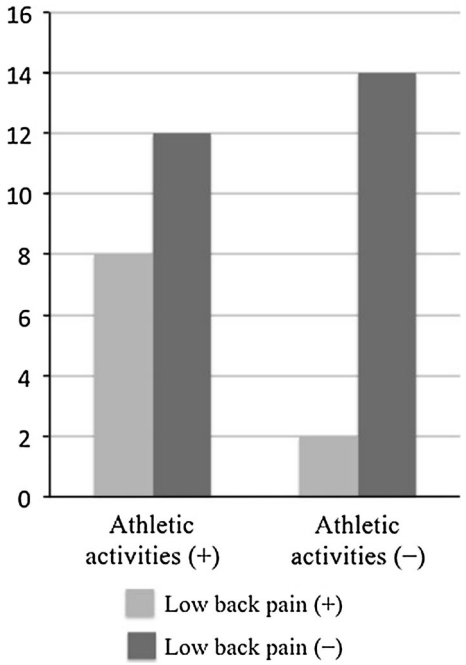

Fig. 2 Proportion of patients who reported low back pain in childhood and adolescent period, according to level of athletic activity

Spina bifida occulta

In 9 of 36 patients (25.0 %), lumbar spondylolysis was accompanied by SBO of the sacrum (Fig. 4); all of them (eight men, one woman) involved the L5 vertebra.

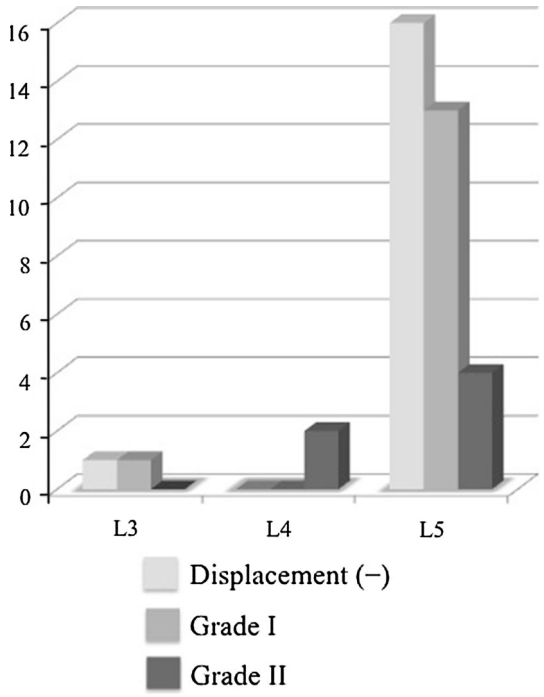

Fig. 3 Proportion of patients with isthmic (spondylolytic) spondylolisthesis and the corresponding lumbar vertebrae distribution. The Meyerding grades of disk displacement are shown. Displacement was seen in 54.1 % of patients (grade I: 14, grade II: 6). In the L5 spondylolysis, 51.2 % had displacement, while in L3 and L4 vertebrae, 75.0 % had displacement

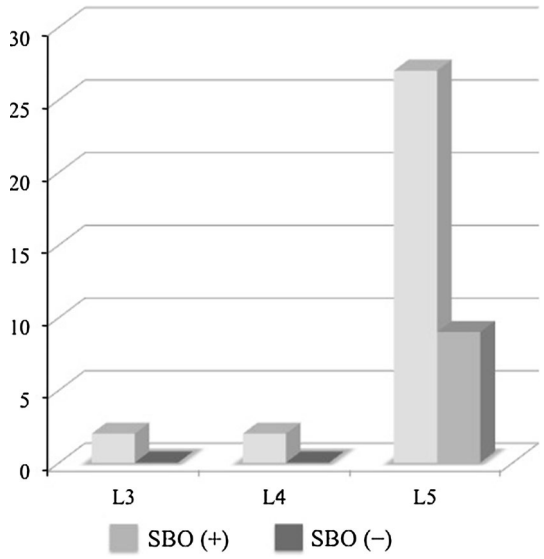

Fig. 4 Proportion of patients with spina bifida occulta (SBO) and the corresponding lumbar vertebrae distribution. In 25.0 % patients, SBO of the sacrum was accompanied by lumbar spondylolysis. All of these patients had L5 spondylolysis

Discussion

Lumbar spondylolysis, as first suggested by Wiltse et al. [1], is present in approximately 6 % of the general population and has been considered as a stress fracture due to its high prevalence in adolescent athletes (20–30 %) [7, 10]. However, sometimes it is insidious and asymptomatic and is diagnosed incidentally. Libson et al. [13] reported a 9.7 % incidence of lumbar spondylolysis based on analysis of plain radiographs of 936 asymptomatic soldiers prior to military placement. In this study, the patients were not diagnosed with lumbar spondylolysis until the average age of 55.8 years (range 25–77 years). Also, approximately 40 % of our study population had unidentified spondylolysis until adulthood and had no history of athletic activity or LBP during their growth period. Thus, both the pathophysiology and role of spondylolysis in causing LBP are still unclear.

In general, spondylolysis does not always evolve into spondylolisthesis. According to past reports, approximately 30–50 % of subjects with spondylolysis never progressed to spondylolisthesis [14–16]. In this study, low-grade (Meyerding grade I or II) spondylolisthesis was found in 54.1 % of patients with spondylolysis, which is similar to the incidence in the general population [10].

In the present study, spondylolysis was accompanied by SBO of the sacrum in 9 (25.0 %) of 36 patients; there was a male predominance, and all of them had spondylolysis at L5. In a recent published report on a cohort of elementary school age children with L5 spondylolysis, 92.6 % had sacral SBO and 59.3 % had SBO involving both the lamina (L5) and sacrum [9]. In addition, there was a report on three boys, including twins, from the same family who had SBO with L5 spondylolysis [8]. These results strongly suggest that male patients with SBO may have some osteogenetic factors that lead to predisposition to spondylolysis.

There have been several papers on the predisposing factors, in addition to stress fracture, in the development of spondylolysis. In the past, some authors reported familial occurrence of spondylolysis [8, 17, 18]. Fredrickson et al. [14] analyzed 23 patients with spondylolysis and their family members and found familial occurrence of 32 % in fathers, 23 % in mothers, and 34 % in male siblings. In reports by Wynne-Davies et al. and Albanese et al. [15, 18], spondylolysis was identified in 19 and 22 %, respectively, of patient's relatives. In addition, Yamada et al. [8] reported three cases of lumbar spondylolysis in juveniles from the same family. These clinical findings suggest a genetic predisposition to lumbar spondylolysis. Recently, Cai et al. [11] have reported results on genetic analysis of dysplastic spondylolysis. They identified a novel

heterogeneous mutation of the sulfate transporter gene in five affected subjects in a Chinese family.

We acknowledge several limitations in this study. For example, we had to rely on only the patients' memory regarding their experiences of LBP in childhood and adolescent period because the information was not available from the official medical records. To resolve this problem, a large prospective cohort investigation that spans many decades would be ideal.

In conclusion, approximately 90 % of patients with terminal-stage spondylolysis that was first diagnosed in adulthood involved the L5. Also, about 40 % had no history of athletic activity or experience of LBP during their growth period. In addition, only some patients with L5 spondylolysis had SBO, and all but one of these patients was male. This suggests that male patients with L5 spondylolysis may have some congenital predisposition.

Compliance with ethical standards

Conflict of interest All authors declare that they have no conflict of interest that could bias the nature of this report.

Ethical standard All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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