Abstract: Three-level lumbar spondylolyses are extremely rare. So far, only 11 cases were reported in the literature. Treatment of multilevel spondylolyses has not been consistent. Conservative treatment is commonly considered first in most patients, but those who remain symptomatic may benefit from operative treatment. We report here 3 cases of 3-level lumbar spondylolyses that were treated successfully with direct isthmic repair in 2 cases and a combined surgery of isthmic repair and interbody fusion in 1 case. Our clinical results indicated that direct defect repair using the screw–hook technique is a simple and safe procedure for the motion segment with normal disc. If the involved disc shows degenerative change, fusion surgery should be considered.

Surgical treatment of multilevel spondylolyses varies between fusion, direct isthmic repair, and combined management associating procedures at different levels. The success of management of the 3 patients with 3-level spondylolyses depends on the choice of appropriate treatment for every patient.

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Abbreviations: CT = computed tomography, MRI = magnetic resonance imaging, ODI = Oswestry Disability Index, VAS = Visual Analog Scale.

INTRODUCTION

Lumbar spondylolysis, a defect in the pars interarticularis, is a common cause of low back pain in adolescents and young adults. It occurs in 6% of the general population and has been reported more commonly in men. Spondylolysis commonly occurs at the fourth and fifth lumbar vertebrae, accounting for more than 95% of total cases of spondylolysis. Lumbar spondylolysis may either occur asymptptomatically or be associated with significant low back pain. Symptomatic pars lesions appear to be particularly a clinical problem in adolescents, especially adolescent athletes. Treatment of spondylolysis has not been consistent. Conservative treatment is commonly considered first in most patients, but those who remain symptomatic may benefit from operative treatment. When the source of back pain is the pars defect itself, direct repair of spondylolysis seems to be most effective and appears to be more logical than posterior spinal fusion in the patients without disc degeneration or spondylolisthesis.

Multilevel spondylolyses are rare, varying between 1.2% and 5.6% of lumbar spondylolysis cases, and majority involve 2 levels, L4 and L5. Three-level lumbar spondylolyses are extremely rare. So far, to our knowledge, only 11 cases of 3-level lumbar spondylolyses have been reported in the existing English literature. In this paper, we reported 3 cases of 3-level lumbar spondylolyses in young adults and review the management choices made in these cases. This study obtained the approval of the medical ethics committee of our hospital (General Hospital of Armed Police Force, Beijing, China). The written informed consent was obtained from the 3 patients before lumbar surgery.

CLINICAL FINDINGS

Case 1, a 22-year-old male, presented with low back pain for 2 years. His back pain was worse after activities, and slightly reduced after bed rest. He did not complain of radicular pain, numbness, and concomitant claudication in his lower extremities. He had a history of lumbar injury dropped down from 2 meters high 2 years ago. Physical examination revealed tenderness and percussion pain over and beside L3–S1 spinous processes. Motor, sensory, and tendon reflex were all normal in his lower limbs. The straight leg-raising test was negative bilaterally. Lumbar radiography and computed tomography (CT) scan showed left L3 and bilateral L4 and L5 spondylolyses without any slippage. Magnetic resonance imaging (MRI) of lumbar spine showed normal discs.

Before admission to our hospital, the patient had undergone a 3-month course of conservative treatment with physiotherapy and nonsteroidal anti-inflammatory drugs in a local clinic, but these therapies did not relieve his low back pain. Because CT scans have showed terminal-stage findings in the isthmic lysis areas, no abnormality of the posterior arches such as dysplasia and spina bifida was found, surgical indication for left L3 and bilateral L4 and L5 direct isthmic repair via a pedicle screw–hook technique was recommended.

Case 2, a 22-year-old male, was admitted with low back pain for 14 months. Pain was originated from an acute lumbar heavy blow 14 months ago, and increased gradually. The activities of daily living were severely limited. One month ago, he felt numbness in his right hip and right lateral thigh. He did not complain of radicular pain in his lower extremities and trouble walking. Physical examination revealed tenderness and percussion pain over L3–S1 spinous processes. Motor, sensory, and tendon reflex were all normal in his lower limbs. The straight leg-raising test was negative bilaterally. Radiographs and CT scans showed left L3–L5 spondylolyses without any slippage (Figures 1 and 2). MRI of lumbar spine showed no sign of disc degeneration (Figure 3).

After his lumbar injury, he was sent to a local hospital, and was diagnosed with L3–L5 bilateral spondylolyses by plain radiography. Since then, he has undergone conservative treatment with bracing to protect lumbar spine, physiotherapy, and oral nonsteroidal anti-inflammatory drugs in a local clinic, but these therapies did not relieve his low back pain. After he underwent a 4-month course of conservative treatment, further...
CT scan showed terminal-stage findings in the L3–L5 bilateral isthmic lysis areas. Surgical indication for bilateral L3–L5 direct isthmic repair via a screw–hook technique was recommended.

Case 3, a 23-year-old man, was admitted with low back pain for 6 months. In the beginning, his back pain can be controlled by oral nonsteroidal anti-inflammatory drugs. A month ago, he sprained his waist carelessly, which aggravated his back pain. Since then, he has undergone conservative treatment in a local hospital, but conservative treatment did not relieve his back pain. He was admitted to our hospital for further treatment. Physical examination revealed a normal lumbar curvature and mild limitation of lumbar activities. Tenderness and percussion pain were obvious over and beside L3-S1 spinous processes. Motor, sensory, and tendon reflex were all normal in his 2 lower limbs. The straight leg-raising test was negative bilaterally. Radiographs and CT scans showed bilateral L3–L5 spondylolyses. T2-weighted MRI showed L5/S1 disc degeneration and normal signal intensity in other lumbar discs.

Because MRI showed L5/S1 disc degeneration, we needed to identify whether this disc was a pain-generating disc. So, lumbar discography was recommended. The patient underwent provocative discography at L4/5 and L5/S1 discs. The result of discography showed L5/S1 disc disruption with pain reproduction, which indicated the L5/S1 disc as the one of the back pain sources. Because CT scan had shown terminal-stage findings in the L3–L5 bilateral isthmic lysis areas, accordingly, surgical indication for bilateral L3 and L4 direct isthmic repair and L5/S1 interbody fusion was recommended.

DIAGNOSTIC FOCUS AND ASSESSMENT
Three cases of 3-level lumbar spondylolyses were diagnosed and subsequently direct isthmic repair in 2 cases and a combined surgery of isthmic repair and interbody fusion in 1 case were recommended for the 3 patients preoperatively based on clinical findings consistent with imaging study.

FIGURE 1. Left sagittal lumbar CT scan showed L3–L5 isthmic lyses. CT = computed tomography.

FIGURE 2. Right sagittal lumbar CT scan showed L3–L5 isthmic lyses. CT = computed tomography.

FIGURE 3. Lumbar T2-weighted MRI showed normal discs. MRI = magnetic resonance imaging.
THERAPEUTIC FOCUS AND ASSESSMENT
Case 1 underwent a surgery of direct isthmic repair in 5 lytic defects. Under general anesthesia, conventional posterior midline approach to lumbar spine was used to exposure L3–L5 spinous processes, vertebral laminae, and facet joints. First, a 6.0 × 45 mm pedicle screw was inserted to involved vertebra under fluoroscopic guidance. Second, the involved lysis area was curetted, freshened, and grafted with autologous iliac bone. Finally, sublaminar hook was placed, and connected through a short rod under compression with corresponding pedicle screw. Subsequently, the remaining repair of 4 lyses was completed in the same way. The total surgical time was 3 hours with 300 mL of blood loss. Postoperatively, the patient remained neurologically intact. A lumbar-sacral brace was used for 3 months after surgery.

Surgical procedure in case 2 was the same as case 1. L3–L5 bilateral lytic defects were repaired with the screw–hook technique (Figure 4). The total surgical time was 3 hours with 500 mL of blood loss. Postoperatively, the symptoms of low back pain and numbness in right hip and right lateral thigh disappeared. A lumbar-sacral brace was used for 3 months after surgery.

Case 3 underwent a combined surgery of isthmic repair and interbody fusion. The surgical procedure was performed in 2 phases. The first phase was L5/S1 interbody fusion. Four pedicle screws were placed in L5 and S1 vertebral bilaterally, then left laminectomy was performed to allow a cage placement (10 × 26 mm) filled with autogenous local bone derived from laminectomy between the decorticated endplates of L5 and S1, and finally 2 rods were connected with 4 pedicle screws under compression. The second phase was L3 and L4 direct isthmic repair bilaterally with the screw–hook technique. The total surgical time was 3.5 hours with 400 mL of blood loss. After surgery, a brace was placed for 3 months, and symptom of low back pain was almost completely disappeared.

FOLLOW-UP AND OUTCOMES
Case 1 has undergone a follow-up of 2 years, and his low back pain was almost completely disappeared. At 6 months after surgery, bone healing was found in all 5 lytic defects. At 8 months after surgery in case 2, bone healing was found in all 6 lytic defects. During a 1-year follow-up period, he was pain-free. At 2-year follow-up in case 3, his low back pain had not recurred, and bone fusion was found between L5 and S1 vertebral bodies and in all 4 isthmi of L3 and L4 according to the evaluation of lumbar radiographs and CT scans.

The surgery for the 3 patients was successfully performed and their symptoms were obviously relieved and lumbar function was markedly improved. The alleviation of back pain and improvement of lumbar function was assessed by the change in the degree of pain with a self-assessment of pain by an 11-point Visual Analog Scale (VAS, 0–10) pain scale and the Oswestry Disability Index (ODI, version 1.0, 0–100). The clinical data and outcomes were summarized in Table 1.

DISCUSSION
Lumbar spondylolysis is a common disease, in more than 90% of cases located in L5.1 However, multilevel isthmic lyses are rare. This is the first report of 3 cases with 3-level spondylolyses.

Pain is the most common initial symptom. Symptoms were correlated to radiographic pathology. A pars defect develops into a chronic nonunion, and becomes bridged by tissue composed of a combination of fibrous, cartilaginous, or osseous material. In this circumstance, the origin of chronic low back pain could be in the scar and connective tissue rich in nociceptive nerve endings that bridges the gap of a pars defect. Extra load exerted on the motion segment may cause disc degeneration, which is therefore an accompanying disorder of spondylolysis. The prevalence of progressive disc degeneration in the involved level is markedly increased in the adult spondylolytic patients who often appear low back pain later in life, after an asymptomatic childhood.

Cause of lumbar spondylolysis is still unclear. In the early 1900s, the pathology of lumbar spondylolysis was considered being congenital failure of fusion of 2 ossification centers or a hyperflexion injury at birth.16 but spondylolysis had never been founded in embryos, fetus and at birth. The earliest patients with spondylolysis have been reported at 6 weeks to 10 months.5 Following Wiltsie’s proposal in 1957,7 the pathology of spondylolysis was shifted to a consensus of fatigue fracture of the pars interarticularis with a strong hereditary basis. For now, 2 factors can explain isthmic lysis, both genetic and mechanical. No specific genetic variation was identified, but it would seem that a genetic predisposition to this pathology may exist. Yamada et al16 reported 3 cases of lumbar spondylolysis in juveniles from the same family. Haukipuro et al15 concluded that inheritance of lumbar spondylolysis is autosomal dominant. Later, Shahriaree et al18 came to a similar conclusion. The incidence varies considerably according to ethnicity and sex.1 The highest incidence was reported in Native residents in Greenland up to 54%.19 From a mechanical point of view, heavy work and repeated injuries seem to favor the appearance of lumbar spondylolysis. A number of studies have described the association of sports activities with spondylolysis, and all of them support the stress fracture theory.1

Radiographs, especially oblique view radiographs are mainstay in the diagnosis of spondylolysis. Traumatic spondylolysis

FIGURE 4. Postoperative 6-month lateral radiograph of lumbar spine.
can be diagnosed early with the use of isotope bone scan or single photon emission computed tomography scan. Preoperative CT scan is very important to evaluate the degree of sclerosis in the bony margins of the isthmic defect. The obvious osteosclerosis was shown in all the isthmic defect margins in the 3 patients by CT scan, which indicated that direct isthmic repair was necessary. Preoperative lumbar MRI is imperative to evaluate the state of intervertebral disc. In our case 3, due to L5/S1 disc degeneration, an additional lumbar discography was used to indicate this disc as a painful disc. This result finally determined the indication for fusion surgery at the L5/S1 segment instead of a direct isthmic repair in L5.

Treatment of multilevel spondylolyses has not been consistent. There are many types of direct repair methods, including Scott’s wiring,20 Buck’s screwing,21 Louis’ butterfly plate,22 and screw–hook technique23 (pedicle screw and hook–rod system). From clinical aspects, screw–hook technique allows for fixation of the defects in the pars interarticularis with rigid implants and exerts force along the lamina to effectively suppress the defect motion, which is critical for better bone healing. The precondition of direct defect repair is normal signal intensity in the involved disc shown in T2-weighted MRI. If not, lumbar fusion should be considered. Although our clinical results are satisfactory, all 3 patients are young and the duration of their follow-up periods has not been long. Therefore, further observation of these patients’ clinical course and radiological results at the segments treated as well as adjacent segments is needed.

### CONCLUSIONS

We reported here 3 cases of 3-level lumbar spondylolyses with good clinical and radiological results. Direct defect repair using the screw–hook technique is a simple and safe procedure for the motion segment with normal disc. If the involved disc shows degenerative change, fusion surgery should be considered. Although our clinical results are satisfactory, all 3 patients are young and the duration of their follow-up periods has not been long. Therefore, further observation of these patients’ clinical course and radiological results at the segments treated as well as adjacent segments is needed.

### REFERENCES

1. Sakai T, Sairyo K, Suzue N, et al. Incidence and etiology of lumbar spondylolisthesis: review of the literature. *J Orthop Sci.* 2010;15:281–288.

2. Saraste H. Spondylolysis and spondylolisthesis. *Acta Orthop Scand Suppl.* 1993;251:84–86.

3. Eisenstein S. Spondylolysis: a skeletal investigation of two population groups. *J Bone Joint Surg Br.* 1978;60:488–494.

4. Nathan H. Spondylolisthesis: its anatomy and mechanism of development. *J Bone Joint Surg Am.* 1959;41:303–320.

5. Witte LL. Etiology of spondylolisthesis. *Clin Orthop.* 1957;10:48–60.

6. Arai T, Sairyo K, Shibuya I, et al. Multilevel direct repair surgery for three-level lumbar spondylolysis. *Case Rep Orthop.* 2013;2013:472968. doi: 10.1155/2013/472968. Epub 2013 Mar 28.

7. Al-Sebai MW, Al-Khawashki H. Spondyloptosis multiple-level spondylolysis. *Eur Spine J.* 1999;8:75–77.

8. Chang JH, Lee CH, Wu SS, Lin LC. Management of multiple level spondylolysis of the lumbar spine in young males: a report of six cases. *J Formos Med Assoc.* 2001;100:497–502.

9. Darnis A, Launay O, Perrin G, Barrey C. Surgical management of multilevel spondylolysis: A case report and review of the literature. *Orthop Traumatol Surg Res.* 2014;100:347–351.

10. Eingorn D, Pizzutillo PD. Pars interarticularis fusion of multiple level lumbar spondylolysis: A case report and review of the literature. *Orthop Traumatol Surg Res.* 2010;69:339–343.

11. Hersh DS, Kim YH, Razi A. Multilevel direct repair surgery for three-level lumbar spondylolisthesis. *Case Rep Orthop.* 2012;2012:472968. doi: 10.1155/2012/472968. Epub 2012 Mar 28.

12. Al-Sebai MW, Al-Khawashki H. Spondyloptosis multiple-level spondylolysis. *Eur Spine J.* 1999;8:75–77.

13. Chang JH, Lee CH, Wu SS, Lin LC. Management of multiple level spondylolysis of the lumbar spine in young males: a report of six cases. *J Formos Med Assoc.* 2001;100:497–502.

14. Nathan H. Spondylolisthesis: its anatomy and mechanism of development. *J Bone Joint Surg Am.* 1959;41:303–320.

15. Sharfi G, Jahanbakhshi A, Daneshpajouh B, Rahimizadeh A. Bilateral three-level lumbar spondylolisthesis repaired by hook-screw technique. *Global Spine J.* 2012;2:51–56.

16. Yamada A, Sairyo K, Shibuya I, et al. Lumbar spondylolisthesis in juveniles from same family: a report of three cases and a review of the literature. *Case Rep Orthop.* 2013;2013:272514. doi: 10.1155/2013/272514. Epub 2013 Sep 26.

17. Haukipuro K, Keränen N, Koivisto E, et al. Familial occurrence of lumbar spondylolysis and spondylolisthesis. *Clin Genet.* 1978;13:471–476.

18. Shahrizaei H, Sajadi K, Rooholamini SA. Family with spondylolisthesis. *J Bone Joint Surg Am.* 1979;61:1256–1258.
19. Simper LB. Spondylolysis in Eskimo skeletons. *Acta Orthop Scand.* 1986;57:78–80.
20. Nicol RO, Scott JHS. Lytic spondylolysis. Repair by wiring. *Spine.* 1986;11:1027–1030.
21. Buck JE. Direct repair of the defect in spondylolisthesis. Preliminary report. *J Bone Joint Surg Br.* 1970;52:432–437.
22. Louis R. Pars interarticularis reconstruction for spondylolysis by plate and screws with grafting without arthrodesis. *Rev Chir Orthop Reparatrice Appar Mot.* 1988;74:549–557.
23. Kakiuchi M. Repair of the defect in spondylolysis. Durable fixation with pedicle screws and laminar hooks. *J Bone Joint Surg Am.* 1997;79:818–825.