Acta Orthop. Belg., 2005, **71**, 750-757

CASE REPORT

High-grade dysplastic spondylolisthesis and spondyloptosis : Report of three cases with surgical treatment and review of the literature

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High-grade dysplastic spondylolisthesis is extremely rare and always involves the L5-S1 level. It is attributed to congenital dysplasia of the superior articular process of the sacrum. It can remain asymptomatic for a long time and can progress to a more severe grade of olisthesis and spondyloptosis. Surgical treatment has varied from posterior-only *in situ* fusion to anterior and posterior fusion with complete reduction. Three cases of symptomatic high-grade (4th and 5th grade) dysplastic spondylolisthesis treated surgically with reduction and fusion are presented. Interbody fusion at the level of olisthesis is crucial.

Key words : dysplastic spondylilisthesis ; spondyloptosis ; surgical treatment.

INTRODUCTION

Dysplastic or congenital spondylolisthesis comprises 15% of the cases of spondylolisthesis presenting before adulthood *(31, 32)*. The articulation between the fifth lumbar and the first sacral vertebra is a site where major dysplasias are encountered. The pars interarticularis may be poorly developed or be elongated. When dysplasia involves both complete absence of the superior facets of the first sacral vertebra and isthmic lysis, then the olisthesis may increase to high-grade slippage.

Dysplastic or congenital spondylolisthesis is found at the L5-S1 level and usually presents in females *(18)*. Genetic predisposition is a very significant causative factor. Patients with dysplastic spondylolisthesis usually develop symptoms during adolescence. Typical clinical symptoms are low back pain and stiffness with hamstring shortening. There is also lumbar hyperlordosis, flexed-hip and -knee walking and toe gait. Radicular pain to the buttocks and thighs is not always present since nerves adapt to tension or compression. Radiation of the pain below the knees or cauda equina syndrome is suggestive of high-grade spondylolisthesis *(26)*.

Despite the wide number of reports on spondylolisthesis in the international literature, there are only few articles commenting on high-grade spondylolisthesis. The latter consists of grade III, IV and V (spondyloptosis) spondylolisthesis as classified by Meyerding *(22)* and comprises 5% of the total cases of spondylolisthesis. Classically, the fifth lumbar vertebra acquires a kyphotic tilt in relation to the sacrum (increased lumbosacral angle, i.e. the angle subtended by the line parallel to the superior endplate of L5 and the S1 endplate

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line), the sacrum acquires a more vertical position (decreased sacral slope, i.e. the angle between the sacral endplate line and the horizontal reference line) and the relative distance of L5 and S1 to the center of the femoral head is changed (sagittal pelvic tilt index, normal value = 1) *(1)*.

Three cases of severe spondylolisthesis-spondyloptosis treated operatively are presented and the literature is reviewed.

CASE DESCRIPTION

Case 1

A 17-year-old adolescent female, a student without any history of strenuous sporting activity, presented with bilateral sciatica. Her past medical history regarding episodes of back pain was negative. Clinical examination showed restriction of forward flexion in the lumbar spine and a straight leg raise test positive at 10º bilaterally. No muscle weakness or sensory disturbances were detected. Plain radiographs showed grade III spondylolisthesis at the L5-S1 level, while computed tomography revealed decreased spinal canal dimensions at L5-S1.

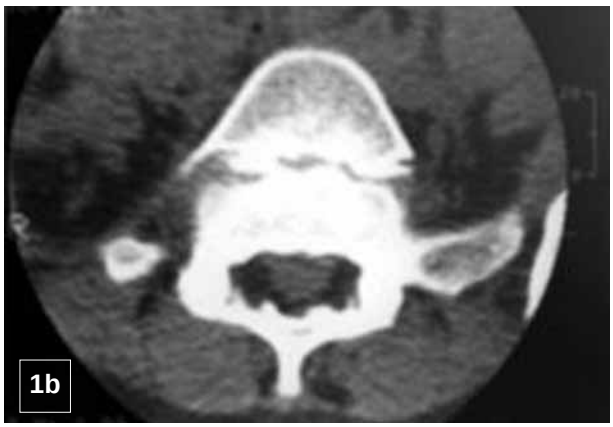

Fig. 1. — **Case 1**-a. Lateral radiograph of the lumbar spine showing grade III spondylolisthesis at L5-S1 ; b. CT of the lumbar spine showing overlapping of vertebral bodies of L5 and S1 ; c. Lateral radiograph of the lumbar spine 6 months postoperatively showing partial reduction and fusion at L5-S1.

Surgical treatment was performed through a posterior approach and included L5 laminectomy, partial reduction of the spondylolisthesis with Harrington distraction rods, sacral dome resection and interbody fusion with corticocancellous graft at L5-S1.

Transpedicular screws connected to rods in compression were used for stabilisation. Postoperatively the patient wore a three contact-point Florida brace for six months. No intraoperative complication was noted and the postoperative course was uneventful. Bone fusion was complete in six months and the patient resumed full activity one year postoperatively (fig. 1). Last follow-up was three years after surgery.

Case 2

A 14-year-old girl presented to our clinic complaining of intense radicular leg pain and numbness in an L5 dermatome distribution bilaterally. Straight leg raise test was positive at 20º bilaterally. No history of low back pain during childhood was reported. Radiographs revealed grade IV anterior olisthesis of the L5 vertebra. Magnetic resonance imaging of the lumbar spine revealed severe degeneration of the L5-S1 disc, but the discs at L3-L4 and L4-L5 also presented mild degenerative changes.

Surgery included L5 laminectomy, decompression of L5 nerve roots bilaterally, partial reduction of the L5 vertebra with distraction. It was not possible to insert screws into the dysplastic L5 pedicles ; in order to achieve anchoring points for distraction-reduction, unilateral screws were inserted at the L3 and L4 level, and posterior L5-S1 fusion was performed with autograft positioned posterolaterally. Postoperatively, a three-point support brace was worn for six months. Three months after removal of the brace, bilateral S1 screw breakage and recurrence of spondylolisthesis was diagnosed. A further surgical procedure was undertaken. The fixation material was first removed through a posterior approach, a new instrumentation was inserted without any reduction attempt, and autologous iliac graft was again applied from the transverse process of L3 down to the sacral ala.

Through an anterior retroperitoneal approach, an interbody cylindrical cage, similar to those used for posterior interbody fusion, was then inserted between the L5 vertebral body and the sacrum. Iliac autograft was positioned between the L5 and S1 vertebral bodies.

Postoperatively, a three-point support brace was used. Two years later, fusion was complete and the patient was symptom free (fig. 2).

Case 3

A 16-year-old female presented with bilateral sciatica and a straight leg raise test positive at 30º bilaterally. Forward bending was restricted in the lumbar spine. Radiology review (plain radiographs and MRI) showed grade V spondylolisthesis-spondyloptosis of L5 on S1.

During surgery, transpedicular screws were first introduced, and then the L5 nerve roots were decompressed bilaterally with hemilaminectomy of L5 and S1 bilaterally. Partial reduction of the olisthesis was done by distraction within the screw-rod system. In the reduction position, titanium mesh cages were inserted in the interbody disc space at L5-S1. Finally, transverse processes decortication and bridging with iliac autograft between L4-S1 was undertaken. Postoperatively, a Florida-type brace was worn for six months, and bone fusion was evident in plain radiographs by the end of that period. The patient reported complete resolution of her symptoms within six weeks. Final follow-up at 3 years showed radiological fusion (fig. 3).

DISCUSSION

High-grade olisthesis and spondyloptosis typically occur in dysplastic type spondylolisthesis. They may also be encountered, but more rarely, in isthmic spondylolisthesis. The dysplastic spondylolisthesis is attributed to dysplasia of the superior sacral articular processes, and consequently it occurs only at the L5-S1 level *(19, 27, 31)*.

Treatment of spondylolisthesis of more than 50% in a growing child is operative *(3)*. As these procedures face major difficulties, many surgical techniques have been described, such as *in situ* fusion, cast reduction and fusion, laminectomy and

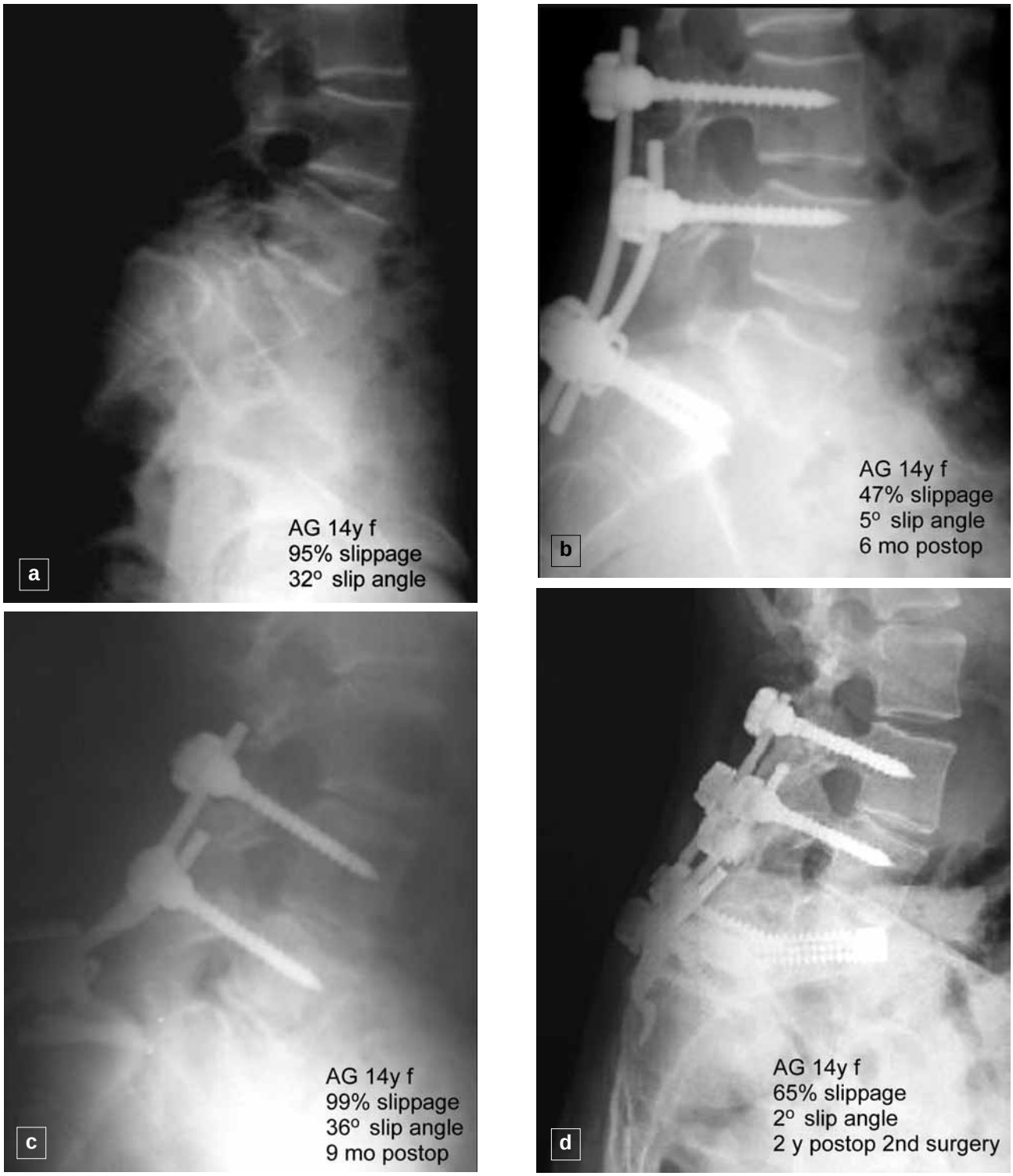

Fig. 2. — **2nd Case** -a. Lateral radiograph of the lumbar spine showing a grade IV spondylolisthesis at L5-S1. Note the vertically positioned sacrum ; b. Lateral view of the lumbar spine showing the instrumentation from L3 to the sacrum. Note that no interbody fusion was performed and there are unilateral screws at L3 and L4 vertebra and no screws at L5 ; c. Nine months postoperatively, lateral radiograph of the lumbar spine showing failure of instrumentation and slip recurrence ; d. Lateral radiograph of the lumbar spine two years following the second surgery, which included revision of instrumentation and interbody cage placement, through an anterior approach.

in situ fusion, reduction and posterior instrumented fusion, reduction and posterior fusion combined with posterior interbody fusion *(8)*, reduction and anteroposterior instrumented fusion with two different exposures, and vertebrectomy (Gaines procedure) *(5)*.

Indications for surgery in high-grade dysplastic spondylolisthesis are mainly related to the following three factors : deformity-kyphosis, instability and progressive deterioration of olisthesis, neurologic deficit *(4, 15)*. In order for a surgical treatment to be effective, it should take these three factors into consideration.

Preoperatively, a detailed diagnostic approach of the disorder with thorough clinical and diagnostic examination is mandatory. The necessary imaging studies of the lumbar spine include plain radiographs, computed tomography and magnetic resonance imaging. In plain radiographs, the grade of olisthesis, the lumbosacral angle as well as the sacral slope should always be measured to predict the deterioration of slippage. Furthermore, the dome-shaped sacrum and trapezoidal shape of the L5 vertebra are also linked to chronic lumbosacral instability *(6)*. Computed tomography reveals the dysplasia of the bone structures and spinal canal dimensions *(21)*. Finally, MRI localises possible pressure on the neural elements and gives information about the disc above the level of olisthesis.

Critical issues still not definitely answered are the necessity of reduction and the degree of advisable reduction, i.e. partial or complete *(2, 17)*. *In situ* posterolateral fusion in children with severe spondylolisthesis was followed by deterioration of olisthesis in 25% of the cases, even though all patients eventually had solid fusion and there were no neurologic deficits *(10)*. There are two studies with long term follow-up of patients fused *in situ* that showed minimal improvement of cosmesis but good clinical results *(14, 28)*. Full reduction may cause neurologic deficit due to compression of the roots during distraction and reduction maneuvers. Also, *in situ* fusion without reduction can be responsible for neurologic symptoms and even cauda equina syndrome *(20)*. The current tendency is to reduce the olisthesis only partially, especially in cases with preexisting neurologic deficits, aiming at restitution of the sagittal alignment and, subsequently, facilitating the bony fusion. The most widespread reduction method for high-grade spondylolisthesis is the one developed by Edwards, i.e. posterior decompression of the neural elements, reduction and fusion with the use of a segmental instrumentation system *(7, 12, 13)*. Combined anterior and posterior procedures are currently performed more frequently for reduction of high-grade spondylolisthesis.

Many studies have been published about the type of fusion. Achieving fusion in severe spondylolisthesis is problematic. The extent of fusion varies with the degree of degeneration of the adjacent discs and the degree of stability provided by the transpedicular screws. However, bone fusion is essential to arrest progression of olisthesis and to preserve a stable mechanical environment. In a study comparing three different types of fusion, i.e. *in situ*, posterior or circumferential (anterior and posterior), the best results were recorded in cases with solid fusion irrespective of the method used *(24)*. In another research paper comparing the results between posterior stabilisation without decompression, posterior stabilisation with decompression and 360º stabilisation, the lowest incidence of pseudoarthrosis was reported in the last category. Moreover, comparison between a group of patients with anterior fusion only and another group with additional transpedicular fixation and decompression in cases of severe spondylolisthesis showed that the group with the added posterior procedure achieved superior results *(25)*. L5 vertebrectomy and reduction of L4 on S1 was described by Gaines for cases of spondyloptosis and has proved to provide excellent fusion rates and improved clinical alignment *(9, 16)*.

Many researchers underline the increased importance of anterior column fusion by reaming across the slip through an anterior or posterior approach, and grafting with a fibula or tricortical iliac graft into the bodies of L5 and S1 without any reduction attempt (Bohlman technique) *(11, 29)*. Anterior column support saves levels, offers better fusion rates and aids in reduction *(23)*. Finally, casting or bracing for the first six months postoperatively probably helps in achieving fusion.

Fig. 3. — **3rd Case.** (a) Lateral radiograph of the lumbar spine showing spondyloptosis of the L5 vertebra. Note the trapezoidal shape of L5 and the dome shaped sacrum ; (b) Sagittal view of lumbar MRI showing degenerative disc changes, severe at L5-S1 and moderate at L4-L5 ; c. Postoperative lateral radiograph of the lumbar spine. Instrumented L4-S1 fusion with interbody mesh cages at L5-S1 and partial reduction of the translation ; d. Three years postoperatively, lateral radiograph of the lumbar spine. Fusion is achieved and the patient is symptom free.

The rate of neurological complications following reduction of high-grade spondylolisthesis is up to 25% *(13)*. These complications include nerve root injury, cauda equina syndrome and injury to the superior hypogastric plexus (causing retrograde ejaculation in males, while in females it does not cause any dysfunction) in anterior procedures. Injuries of the higher lumbar roots have even been reported following reduction of spondylolisthesis at L5-S1 *(30)*. Meticulous surgical technique, nerve root decompression before reduction and reduction of the translation are required in order to reduce the complication rate *(5)*.

In the presented cases, posterior decompression of the neural elements, partial reduction of the olisthesis, stabilisation with transpedicular fixation systems and interbody and posterolateral fusion was performed. The pseudoarthrosis and screw breakage in the second case resulted from inadequate support of the lumbosacral junction in the reduced position due to insufficient anchoring points cephalad to the olisthetic level and non-performance of interbody fusion during the initial surgery. Spinal fusion was eventually achieved in all three patients and their symptoms were cleared. The infrequency of high-grade spondylolisthesis - spondyloptosis and the high risk of postoperative complications necessitate superior preoperative planning, meticulous surgical technique and close postoperative monitoring of the patient. These will lead to a successful treatment outcome.

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