A CLINICAL SYNDROME ASSOCIATED WITH SEVERE LUMBO-SACRAL SUBLUXATION*


The term subluxation has been used in this title because it denotes forward displacement of the last lumbar vertebra on the sacrum without a defect of the interarticular part of the neural arch. Some authorities accept the term spondylolisthesis only when a neural arch defect is present, and describe those cases without such a lesion as subluxation despite the marked attenuation of the neural arch and the giving way of the lumbo-sacral joints. Another term, spondyloptosis (Neugebauer 1888), has also been used to describe the more severe type of displacement. The more extensive the study of the literature the greater becomes the confusion in the distinction between spondylolisthesis and subluxation, and eventually the conclusion must be that spondylolisthesis means nothing more than "a slipping vertebra," as its name implies and as Kilian originally described it in 1854, and that separation into spondylolisthesis and subluxation is false.

In the study of a series of 319 cases of spondylolisthesis (Newman 1963) five groups were described. These groups were defined on the basis of clinical and radiological observations and of the findings at operation. Lumbo-sacral subluxation, of which the two main causative factors are insufficiency of the superior sacral facets and attenuation of the neural arch, was selected as Group 1 (Figs. 1 and 2). In this group there was often no break in the pars interarticularis and yet the degree of slip was apt to be severe (Fig. 3); displacement was seen earlier in life than in the other groups, and aplasia of the neural arches of the upper part of the sacrum was always present. Clinically it was noted that patients with a severe degree of

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slip often presented a characteristic syndrome of symptoms and signs. The least degree of slip that gave rise to the syndrome was a displacement by half the antero-posterior diameter of the body of the first sacral vertebra. It seemed relevant to include in this study all patients with this degree of slip or more, so as to expose the frequency of the syndrome in relation to severe displacement.

Thirty-four patients came into this category. A graph shows the age at onset of the first symptoms (Fig. 4). Twenty-nine of the thirty-four patients began to notice symptoms between the ages of nine and nineteen; the average age was just over fourteen years. The typical syndrome was seen in twenty-five of the twenty-nine patients with onset of symptoms between these ages but it was not observed in any of the five patients whose symptoms began when they were over twenty. This observation suggests that the physiological changes which occur around the age of puberty promote or aggravate the syndrome. This view receives support from the fact that the average age at onset of symptoms (13.0 years) in the female is nearly two and a half years less than that in the male (15.4 years). It is interesting that the onset of symptoms in slipping of the upper femoral epiphysis has a very similar distribution, with girls about two and a half years ahead of boys (Jerre 1950) (Fig. 5).

THE CLINICAL SYNDROME

The presenting symptom was pain, stiffness or deformity, or any combination of them. Pain—Pain was felt in the lower back, buttocks, thighs, and sometimes in the legs, feet and toes. There were two distinct types of pain: a dull ache in the low back, buttocks and thighs made worse by standing and bending and various postural stresses; and a more severe pain radiating to the legs and feet, burning or lancinating in type and made worse by coughing and sneezing and associated in some cases with signs of involvement of the roots or cauda equina.
Stiffness of the spine—Stiffness was the outstanding feature. A patient previously able to touch the toes was often unable to reach farther than the lower thigh or patellae with the finger tips; in severe cases there was virtually no flexion either of the lumbar spine or of the hips (Figs. 6 and 7). The hamstrings were held in spasm. Tightness of the hamstrings, associated with spondylolisthesis, has been reported by Phalen and Dickson (1961), and the two patients whom they described, one a boy of twelve and the other a girl of ten years, had the typical features of this clinical syndrome.
Deformity and gait—The spine often shows scoliosis (Figs. 8 and 9). Commonly it is a list to either side like that in sciatica, but less often there is a definite curve characteristic of the idiopathic structural type. Alternating scoliosis was seen in two cases.

The patient walks with an odd gait typical of a space-occupying lesion involving the cauda equina. It has been well described as a “pelvic waddle”; the knees are slightly bent, the lumbar spine is stiff and there is a marked lordosis with a backward tilt of the pelvis and a crease in the abdomen (Fig. 10). The hips also are held stiffly. The base for walking is wide.
Postural features. Figures 11 and 12—Erect posture, showing backward tilt of pelvis. Figure 13—Restriction of straight leg raising.

Myelography. Antero-posterior and (right) lateral views showing complete block at the level of the neural arch of the fifth lumbar vertebra.
and there is a tendency to swing one or both legs and to walk on the toes. Such a gait may be expected to cause the least possible excursion of the roots and cauda equina. A rather similar gait has been described in the "filum terminale" syndrome (Garceau 1952, Johnson 1952).

Another characteristic feature is that the forward inclination of the pelvis is reduced. In a normal person the pelvis is tilted forward so that a line drawn circumferentially from the posterior superior spine to the symphysis pubis is inclined downwards. The average inclination is 31 degrees in the male and 28 degrees in the female (Wiles 1937). In these patients this line may be horizontal (Figs. 11 and 12).

Straight leg raising, with the patient supine, is much limited, often without pain, and dorsiflexion of the foot with the limb resting on the couch is often restricted (Fig. 13). Neurological phenomena—Pain and paraesthesiae radiating into the leg were common; hot lancinating pain made worse by coughing or sneezing was slightly less common. One or both ankle jerks were absent or diminished in seven cases and the bladder was affected in five instances. Loss of sensation was found on six occasions but no muscle weakness was recorded.

Radiography—Radiographic investigation reveals the extensive lumbo-sacral slip. The body of the last lumbar vertebra slips forward across the upper surface of the sacrum and in severe instances may roll down the pelvic surface of the first sacral vertebra. Other typical abnormalities may be seen in the radiograph (Newman 1963): 1) attenuated neural arch of fifth lumbar vertebra; 2) subluxated lumbo-sacral zygapophysial joints; 3) absence of sacral ridge.

Myelography—Myelography was not performed as a routine; it was done in only six cases in this series. In three there was a complete block and in three a partial block. In each instance the site of block was above the sacrum at the level of the neural arch of the fifth lumbar vertebra (Fig. 14).

A study of the radiographs in this series shows how easily the cauda equina, held anteriorly by the laminae of the fourth and fifth lumbar vertebrae and posteriorly by the body of the sacrum, may be stretched and compressed (Fig. 15).

The distance between the anterior edge of the lamina of the fifth lumbar vertebra and the superior angle of the first sacral body will increase gradually as the slip proceeds, but if trauma or laxity of the ligaments caused by hormonal influences such as occur at puberty or during pregnancy allows further slip, symptoms may be precipitated. The estimated distance between these two points ranges from three-quarters of an inch to two inches in minor and major degrees of slip.

Effect of severe degree of slip on pregnancy and labour—Difficulty with labour is often expected in severe spondylolisthesis and it is not unknown for patients to be warned that Caesarean section will be necessary or even that it is unwise to become pregnant.

Seven of these patients had fifteen pregnancies; twelve deliveries were normal and three delayed, one requiring forceps. Caesarean section was not necessary (Fig. 16).

TREATMENT

In the earlier cases an attempt was made to reduce the degree of forward slip before spinal grafting. Skin traction was applied to both legs and the pelvis was suspended in a canvas sling hung from two Balkan beams. It was thought that some degree of reduction could be achieved and maintained permanently. In the author's experience, although a moderate
degree of reduction was possible in a few instances, it was never maintained during the period of consolidation of the graft. In three instances disturbance of bladder function was produced during traction: in two bladder function returned to normal with release of traction but in the third case there was no recovery. Paraesthesiae in the feet and in one case transient drop foot also occurred during traction.

Since these experiences, attempted reduction by traction or by any other method has not only been abandoned but has been regarded as dangerous.

**Treatment by operation**—Operation is thought to be indicated in two groups of patients: 1) in the adolescent with a degree of slip of half a diameter or more; and 2) in the adult with severe, persistent pain, especially in the presence of neurological signs.

**Surgical stabilisation**—Whether or not they exhibit the typical clinical syndrome, adolescents with mild or severe symptoms who radiologically have a severe degree of subluxation are best treated by bone grafting. The reasons for advising a stabilising operation at this age are clear: 1) to prevent forward subluxation which has a tendency to increase; 2) to relieve either type of pain; 3) to relieve muscle spasm causing stiff back and tight hamstring muscles; and 4) to avoid the danger of tension on roots and cauda equina.

In adults a stabilising operation is less often necessary, but apart from the obvious reason for treating persistent pain there is still the possibility of progressive subluxation, especially during pregnancy.

**Technique of operation**—Stabilisation by bone grafting the lower two lumbar segments to the sacrum is not usually a difficult or a dangerous operation but in this particular condition it is more hazardous. At least part of the sacral roof is aplastic, being represented by a fibrous sheet. Unless it is recognised at the outset that there is very little protection for the dura in the sacrum, damage can be done.

The lack of bone on the sacrum and the deep suprasacral pit produced by the displacement makes bone grafting, even with small slivers, a rather precarious step. It is probably unwise to attempt to interfere with the zygoapophysial joints, poised as they are at such an angle, but preferable to surround them with bone as they stand, lest further subluxation occur.
These points are borne in mind while the back of the sacrum and the neural arches of the fourth and fifth lumbar vertebrae are exposed and cleaned. The spine of the fifth lumbar vertebra may be found indenting the fibrous sacral roof; it is quite stable, not mobile as in the common type of spondylolisthesis. The inferior facets of the fifth lumbar vertebra will have slipped away from the superior sacral facets and the lamina of this vertebra will be found extending down into the depths of the suprasacral pit. As much as possible is made of the bone available on the sacrum and the lowest two lumbar laminae. The muscles on each side are now reflected still further laterally to expose the lateral masses of the sacrum. Further proximally the transverse processes of the fourth and fifth lumbar vertebrae are exposed. The transverse processes, the lateral masses and the lateral and posterior aspects of the facets are decorticated. Multiple slivers of iliac bone are fitted into place. Those packed down towards the transverse processes lie in a tunnel between bone and muscle and make a valuable addition to the posterior fusion (Fig. 17).

In addition to fusion a preliminary decompression was performed on four occasions; in both patients with bladder paralysis, in one adult with paraesthesiae in both feet and in another patient in whom myelography showed a complete block. Obstructing bone from the fourth and fifth laminae was removed in each case.

*Fig. 18—Scoliotic list before (left) and after operation.*
*Fig. 19—Range of forward flexion before (left) and after operation.*
*Fig. 20—Range of straight leg raising before (left) and after operation.*
Management after operation—The patient remains in a plaster bed for eleven weeks after operation. During the twelfth week he is allowed free in bed and later is encouraged to stand upright. When the upright position can be maintained for fifteen minutes a spinal brace is fitted. A Gauvain brace has been found to be the most suitable. It is worn for six weeks; thereafter spinal exercises are encouraged with the object of regaining full spinal movement above the fused segments within six months of operation (Figs. 18 to 20).

RESULTS

Thirty of the thirty-four patients underwent stabilising operations. The four not treated by operation complain of continued pain; three had stiffness and two are unable to do their usual work.

Of the thirty treated by operation, only two complain of pain and none of stiffness; all are following their usual occupation. Two have incontinence of urine; in one this was produced during traction and it did not recover when the traction was released nor after subsequent decompression and fusion. The other patient had been incontinent since a previous operation. It was thought that displacement had been increased during operation, possibly from removal of holding bone. Spinal list disappeared in all cases. In one case an idiopathic scoliosis progressed and stabilisation at a higher level was necessary.

Fusion appeared radiologically sound in all cases. A fusion rate of 100 per cent for posterior lumbo-sacral grafting in adults would be an exceptional claim, but in this series all operations except two were done on patients under the age of twenty and in each instance the patient remained in a plaster bed for three months. One can assume that at this age stabilisation can quite easily be achieved.

Effect of fusion on progressive slip—The degree of slip was measured by the relation of the postero-inferior angle of the body of the fifth lumbar vertebra to the superior and anterior surface of the first sacral body. One line was drawn across the superior surface and another down the anterior surface and each was divided into ten equal units (Fig. 21). Measurements were taken in twenty-four cases followed long enough for adequate comparison. Increase in slip was noted in nine instances (Figs. 22 and 23). In six patients in whom the spine
was not stabilised or in whom increase in slip was measured before stabilisation there was a total of twenty-eight years' observation and seventeen units of slip were found. In eighteen patients with stabilised spines there was a total of 105 years' observation and four units of slip were found. Thus, on average, an unstabilised spine showed one unit of slip in 1.6 years, whereas a stabilised spine showed one unit of slip in 26 years.

SUMMARY

1. Thirty-four patients with severe lumbo-sacral subluxation have been studied. Twenty-nine of these came for advice between the ages of nine and nineteen, and of these, twenty-five developed symptoms and signs of a characteristic syndrome.

2. The details of the syndrome are described: the essential features are spinal stiffness, a lordotic gait, resistance to straight leg raising, and in some cases evidence of interference with cauda equina or nerve root.

3. The danger of attempted reduction by traction is stressed, as well as the difficulties to be encountered during posterior lumbo-sacral fusion.

4. The reasons for operating are given; the results of spinal fusion were satisfactory.

5. The traditional apprehension concerning the effect of severe subluxation on childbirth has probably been over-stressed.

6. The tendency to slip was almost completely arrested by spinal fusion.

REFERENCES


