The Natural History of Spondylolysis and Spondylolisthesis

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ABSTRACT: **We performed a prospective roentgenographic study to determine the incidence of spondylolysis, spondylolisthesis, or both, in 500 unselected first-grade children from 1955 through 1957. The families of the children with spondylolysis were followed in a similar manner. The incidence of spondylolysis at the age of six years was 4.4 per cent and increased to 6 per cent in adulthood. The degree of spondylolisthesis was as much as 28 per cent, and progression of the olisthesis was unusual. The data support the hypothesis that the spondylolytic defect is the result of a defect in the cartilaginous anlage of a vertebra. There is a hereditary predisposition to the defect and a strong association with spina bifida occulta. Progression of a slip was unlikely after adolescence and the slip was never symptomatic in the population that we studied.**

The classification and etiology of spondylolisthesis have been discussed at great length[1,38,43,44]. In this study we attempted to delineate the natural history of isthmic spondylolysis and spondylolisthesis in childhood and adolescence. Isthmic spondylolisthesis has been defined as a condition in which "fibrous defects are present in the pars interarticularis which permit forward displacement of the upper vertebrae and separation of the anterior aspects of the vertebra from its neural arch"[1]. The etiology of the bone defect has been debated for many decades. The earliest recorded theory is that of Rambaud and Renault[24] in 1864. They related the defect to failure of fusion of two separate ossification centers. Willis, in 1931, disagreed with this hypothesis, suggesting instead that the defect was the result of anomalous ossification, and that trauma played a secondary role[37]. Hitchcock, working with infant cadaver spines, suggested that hyperflexion at the time of delivery was the cause of the defect and that healing of these fractures was prevented by the marked vascularity of the area[11]. Rowe and Roche reported on the dissection of the spines of 500 infant cadavera in 1953 and found no neural-arch defects[24]. That discredited Hitchcock's theory of hyperflexion at the time of birth and lent support to Willis' hypothesis.

Wiltse theorized in 1957 "that the lesion in the pars interarticularis results from dissolution of continuity of the bone due to a congenital weakness at this point"[40]. He went on to say that the defect was of varying expressivity, and that the exact nature of the congenital weakness permitting dissolution remained an enigma. More recent authors have favored Wiltse's theory.

The natural history of isthmic spondylolysis and spondylolisthesis has also been studied and debated for many years. While reports of their incidence in adults are available[3,7,8,10,13,21,23,28,31,37] and the fetal incidence has been shown conclusively to be zero[4,8,11,24], there has been no report, retrospective or prospective, that described the natural history of these entities to adulthood. There have been many reports of children who were found to have a pars interarticularis defect on roentgenograms[25,26,34,36], but these reports unfortunately failed to delineate the course of asymptomatic children with spondylolysis, spondylolisthesis, or both, since they concerned mostly isolated cases of patients who were seen because of pain or deformity, or both. Friberg[8] recognized this lack of data and stated: "In order to judge the average frequency, it is not anatomic or clinical studies that are required, but investigations of a slice of normal population, for example a group of military conscripts or school children."

We agree with Friberg that such a study was needed to answer the following questions: (1) What is the incidence of spondylolysis and spondylolisthesis in childhood, adolescence, and adulthood? (2) Is the development of a defect painful? (3) When does slipping occur, and is it painful? (4) Are there factors that are useful to prognosticate whether or when a patient will have an increase in the percentage of slip? (5) Is there evidence of genetic transmission?

Materials and Methods

Our main study group consisted of 500 first-grade children. The only criterion for inclusion in the study was that the child attend any one of several local public elementary schools in the years 1954 to 1957. Parental permission was obtained for each child in the study, which was conducted in a relatively small community on the border between New York and Pennsylvania. Most of the children in each class participated. The small size of each class made it necessary to include classes in three consecutive years in order to obtain an adequate population for the study.

Supine anteroposterior, lateral, and oblique roentgenograms of the lumbar spine were made for each child. The children were also checked clinically for any spinal deformities. The roentgenograms and all ancillary studies were

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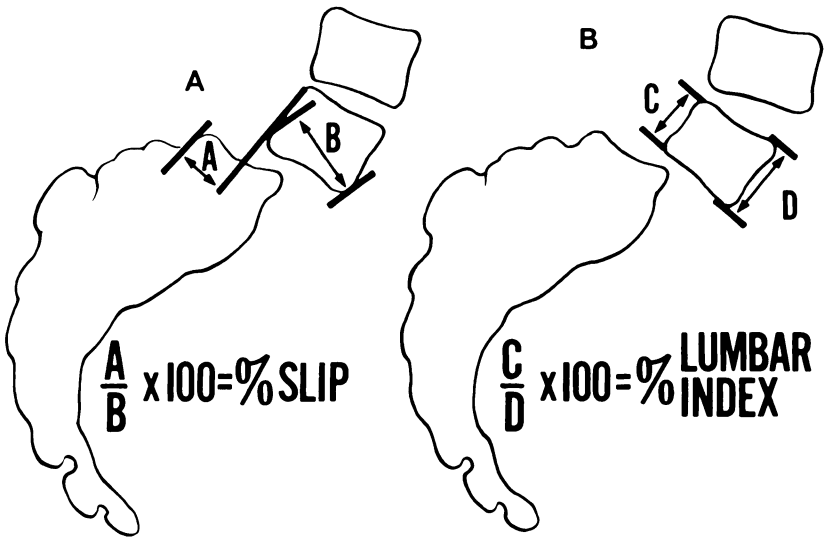

FIG. 1

A, The method of measuring the degree of vertebral slip as modified by Taillard[12]. *B*, The method of calculating the lumbar index.

reviewed by us and a radiologist. Any questionable defect was studied with supplemental oblique roentgenograms. Tomography was used when further clarification was needed. When the existence of a defect was questionable, the roentgenograms were repeated one year later. All subjects available for follow-up had repeat roentgenograms at the ages of ten to twelve, fifteen to sixteen, and eighteen years or older. Children who were not readily available for follow-up were traced through school and hospital records whenever possible. The final extensive search for missing patients was carried out from 1977 to 1979 using school and hospital records. Some patients were lost to follow-up (Figs. 2 and 3) because the family moved out of state or because the parents or child refused to have any more roentgenograms made. The same radiologic technician made all of the roentgenograms. The study was terminated in 1979 after twenty-five years, at which time more than 1500 sets of roentgenograms had been made and further attempts at locating patients had been unsuccessful.

Three ancillary studies were conducted at the same time as the main study. The first ancillary study involved twenty-three of the families of children from the primary study who had a pars interarticularis defect on the initial roentgenograms. Similar roentgenograms were made for every family member who was more than one year old. These families, including any children born subsequently to any family members, were followed until 1979.

In the second ancillary study we reviewed the roentgenograms of 500 normal neonates from the same community. Anteroposterior, lateral, and oblique roentgenograms were available for each child. The pars interarticularis is cartilaginous in the neonate, so a roentgenographic diagnosis of spondylolysis could not be made; however, spondylolisthesis, if present, could be demonstrated.

The third ancillary study involved thirty-two consecutive families. The only criterion for inclusion in the study group was that the mother must have delivered a child in the community's obstetrical unit in 1955 or 1956 and agreed

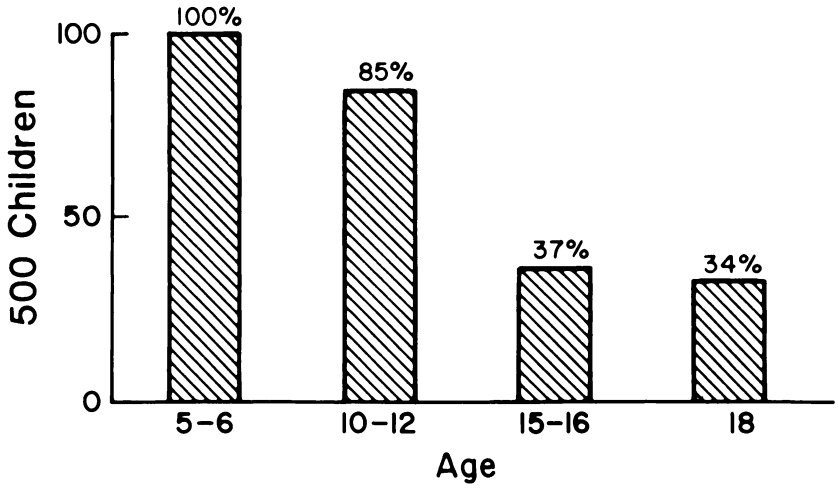

FIG. 2

Bar graph showing the per cent of patients available for follow-up examinations.

to have standard roentgenograms made for all family members. We intended this study to provide a comparison with the first ancillary study. The parents, index neonate, and all siblings were examined. Subsequent roentgenograms of the neonate were made at the ages of one, two, and three years.

We determined the presence of spondylolysis with or without spondylolisthesis in each subject by consensus, only after repeated review of all the roentgenograms. A child was considered to have spondylolysis, as defined in The American Academy of Orthopaedic Surgeons' *A Glossary on Spinal Terminology*[1], if a unilateral or bilateral defect in the pars interarticularis could be identified. Isthmic spondylolisthesis, as defined in the *Glossary*[1], was measured by a modification of Taillard's method[32]. Sacralization or lumbarization was recorded, considering the first vertebra that was not rib-bearing to be the first lumbar vertebra. Absence of a spinous process or lamina, or both, on the anteroposterior roentgenogram was recorded as spina bifida occulta, and we noted whether the spina bifida was confined to the sacrum or if it extended into the lumbar region. We also recorded the lumbar index (Fig. 1, *B*), the sacral inclination, and the lumbar lordosis for all patients in the first study group of 500 children.

The data obtained from the first ancillary study were analyzed by segregation analysis for the possibility of genetic transmission[18]. Family pedigrees were extracted from the literature[10,21,26,39,40] and subjected to a similar analysis. The family pedigrees from the first ancillary study and from the literature were then combined and analyzed using the same technique.

Results

In the main study, of the 500 children who were first examined while they were in first grade, follow-up roentgenograms were made for 85 per cent in the ten to twelve-year-old age group, 37 per cent in the fifteen to sixteen-year-old group, and 34 per cent in the eighteen and older age group (Fig. 2). The male:female ratio was 49:51. In this group, 116 subjects did not have a defect of the pars interarticularis and had complete sets of roentgenograms made at each routine follow-up visit. These 116 subjects provided a base for comparison with the patients found to have a pars interarticularis defect.

Twenty-two children (4.4 per cent of the original 500) were found to have a unilateral or bilateral pars interarticularis defect of the lumbar spine at the age of six years. Nineteen of these defects, twelve in boys and seven in girls, were at the fifth lumbar vertebra; one, at the first; one, at the second; and one, at the fourth (Fig. 3). Of the pars interarticularis defects at the fifth lumbar vertebra, fifteen were bilateral and four were unilateral. The defects at the first, second, and fourth lumbar vertebrae were all unilateral. Subsequent roentgenograms revealed that eight additional patients had a defect at the fifth lumbar vertebra: in two girls and one boy at the age of twelve years, in one boy at the age of fourteen, in two boys at the age of sixteen, and in two men more than eighteen years old. One male and one female patient had a unilateral defect only; the remaining six had a defect bilaterally when first discovered. This came to a total of thirty patients with spondylolysis, an incidence of 6 per cent. In twenty-seven of these (eighteen male and nine female patients), the defect occurred at the fifth lumbar vertebra, an incidence of 5.4 per cent. Healing of the defect was demonstrated in only one patient, a six-year-old girl with a unilateral defect at the fifth lumbar vertebra. The roentgenograms of this patient at the ages of ten and twelve demonstrated persistence of the defect. It had healed by final follow-up when the patient was twenty-eight years old. At final follow-up, twenty-one patients had a bilateral defect at the fifth lumbar vertebra and five had a unilateral defect there. A defect developed bilaterally in two patients after the initial roentgenographic examination had revealed a unilateral defect. No patient with a pars interarticularis defect was lost to follow-up once the defect was discovered.

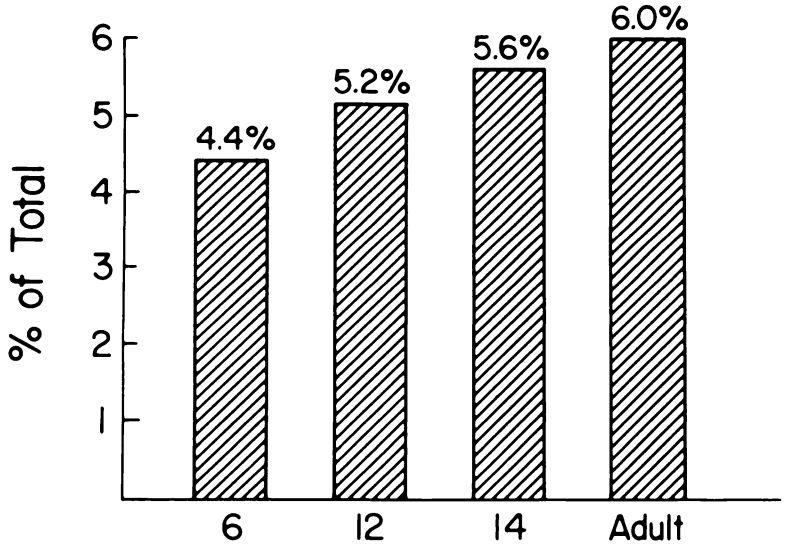

FIG. 3

Bar graph showing the incidence of spondylolysis by age.

The information we report in the remainder of this paper regarding spondylolysis, spondylolisthesis, sacralization, lumbarization, spina bifida occulta, and lumbar index is based mostly on the cases of the 116 patients (sixty-one male and fifty-five female) who did not have a pars interarticularis defect and on the cases of the twenty-seven patients who were found to have a defect at the fifth lumbar vertebra.

Spondylolisthesis was seen in thirteen of the nineteen patients in whom spondylolysis had been ascertained by the initial roentgenograms of the 500 children. Spondylolisthesis was seen in twenty of the twenty-seven adults with a pars interarticularis defect. The over-all amount of the slip ranged from zero to 30 per cent. The average increase in slip was 16 per cent in male patients and 14 per cent in female patients. Spondyloptosis was not seen in this series. Four male patients who had a defect at the age of six had an increase in the over-all slip of 10 to 28 per cent. The serial roentgenograms of these four patients showed narrowing of the disc space between the fifth lumbar and the first sacral vertebra. The severity of narrowing coincided with the increase in olisthesis. No female patients who had a defect at the age of six had an increase in the slip of more than 10 per cent. Slipping was noted to occur during both decades under study, with the largest change occurring during the early teen-age years. No slip increased after the patient reached the age of eighteen. Three women were examined roentgenographically five to six years post partum and there was no evidence of an increase in the slip.

Three of the eight children who were initially normal but who had development of a defect on follow-up roentgenograms had a slip of at least 10 per cent on the first roentgenogram that showed the lesion (Fig. 4). Two were boys with a 10 per cent slip, and one was a girl with a 17 per cent slip. No increase in slip was noted in any of them.

The three patients with spondylolysis not involving the fifth lumbar vertebra had a unilateral defect and never showed a slip. No slip developed in seven of the patients with a defect at the fifth lumbar vertebra. Three of these seven had a bilateral defect and four had a unilateral defect. No patient with a unilateral defect had development of a slip unless a defect also developed in the opposite pars interarticularis.

The roentgenographic appearance or absence of spina bifida occulta is dependent on age. The data referring to spina bifida occulta are therefore given by age group. Fifty-nine per cent of the 116 subjects who were followed to maturity and who did not have a pars interarticularis defect had spina bifida occulta at the lumbosacral level at the age of six. Only 18 per cent of the same group had spina bifida occulta that involved the fifth lumbar vertebra. Only 20 per cent of the 116 subjects had some form of spina bifida occulta as adults, and a defect involving the fifth lumbar vertebra could be found in only 7 per cent of the total group. In addition, spina bifida occulta at one or two levels, identified on roentgenograms in childhood, ossified completely by adulthood in most subjects, whereas defects involving three or more levels were only partially ossified by adulthood.

The over-all incidence of spina bifida occulta in patients with a pars interarticularis defect was 92 per cent in childhood, and 55 per cent had spina bifida occulta involving the fifth lumbar vertebra. Spina bifida occulta persisted into adulthood in 70 per cent of patients with a pars interarticularis defect. Thirty per cent continued to have roentgenographic evidence of spina bifida occulta involving the fifth lumbar vertebra.

Nine of the 116 normal subjects had some degree of

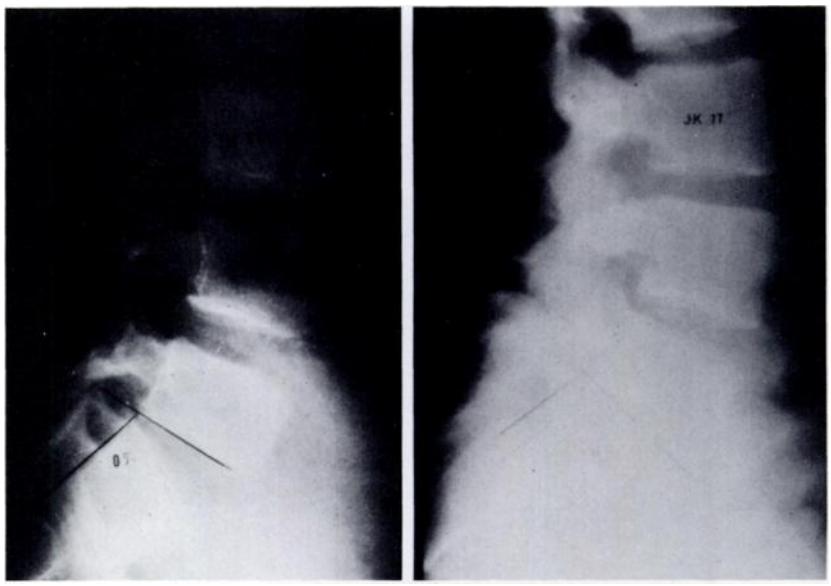

FIG. 4-A FIG. 4-B

Fig. 4-A: Roentgenogram of a ten-year-old patient, showing no evidence of spondylolysis or spondylolisthesis.
Fig. 4-B: Roentgenogram of the same patient at the age of twenty-seven, showing a bilateral pars interarticularis defect at the fifth lumbar vertebra and a slip of 10 per cent.

sacralization and nine others had some lumbarization, for an over-all incidence of 16 per cent. In the group of twenty-seven patients with a pars interarticularis defect at the fifth lumbar vertebra, one patient had lumbarization and one had sacralization, for an over-all incidence of 7 per cent. This difference (16 per cent compared with 7 per cent) is not statistically significant.

The lumbar index was tabulated in each age bracket for both the 116 normal subjects and for the group with spondylolysis. The normal subjects had average values of 80, 84, and 85 per cent in childhood, adolescence, and adulthood, respectively. There was no significant difference between male and female subjects. The lumbar indices in the patients with spondylolysis averaged 78, 78, and 73 per cent. There was a significant difference between the adult male and female patients: 69 per cent compared with 79 per cent. This difference in the lumbar index corresponded to a difference in the percentage of slip: 17 per cent in men compared with 14 per cent in women. A higher percentage of slip was related in either sex to a lower lumbar index.

The children with a known pars interarticularis defect were closely followed. The parents were instructed to notify us and bring the child in for examination whenever any symptoms developed referable to the spine or lower extremities. In addition, we searched the hospital emergency-room records of these children for any admission data involving low-back pain. Low-back pain was found to have developed in four patients during the twenty-five years of follow-up. One female patient, in whom a bilateral defect had developed at the fifth lumbar vertebra when she was twelve years old, had low-back pain and right radicular symptoms when she was sixteen years old. The pain and symptoms resolved with conservative care until she was twenty-eight years old, when they recurred. One male patient, found to have a unilateral defect at the fifth lumbar vertebra at the age of fourteen, began having symptoms of herniation of the disc between the fourth and fifth lumbar vertebrae at the age of twenty-three and underwent a laminectomy and discectomy. One male patient who was not known to have a pars interarticularis defect had a single episode of low-back pain at the age of twenty-two, at which time roentgenograms revealed a bilateral defect at the fifth lumbar vertebra. Low-back pain and radicular symptoms developed in a twenty-seven-year-old woman who had bilateral defects on her initial examination. The symptoms resolved with conservative treatment.

Twenty-three families in the first ancillary group were available for study. Seven of the twenty-two fathers (one had died), or 32 per cent, were found to have spondylolysis. Four (17 per cent) of the twenty-three mothers had similar findings. Sixteen (34 per cent) of the forty-seven male siblings also were found to have spondylolysis. Segregation analysis was used to study the possibility of genetic transmission in this group. This form of analysis tests various hypothetical modes of genetic inheritance and compares the possibility of recurrence of each hypothesis with that actually observed. Several different hypotheses were tested. The two most likely hypotheses were: (1) mating of a normal man and woman with mendelian recessive inheritance of the defect, or (2) mating of a normal person and a person with a spondylolytic defect with mendelian dominant inheritance. The possibility of the first hypothesis occurring, according to our data, was not rejected, but there was evidence of either reduced penetrance or the presence of sporadic or non-inheritable cases. The second hypothesis also was rejected, but the data indicated the possibility of reduced penetrance.

In the second ancillary study we reviewed the roentgenograms of 500 newborns. There was no evidence of spondylolisthesis at any level in any of the roentgenograms.

The third ancillary study involved thirty-two mothers, their newborns, and their families. Three parents, two fathers, and one mother were found to have a pars interarticularis defect, an incidence of 5 per cent. Sixty-nine siblings, ranging from one to twenty-four years old, were examined at least once. One (7 per cent) of the fifteen children in the three families in which one parent was found to have a pars interarticularis defect had spondylolysis. No children were found to have spondylolysis in the remaining families whose parents were unaffected. None of the newborns had spondylolisthesis, and sequential roentgenograms made at the ages of one and two again did not show evidence of spondylolysis. Eight of the original thirty-two newborns were examined roentgenographically in adulthood, and none were found to have a pars interarticularis defect.

Discussion

The natural history of isthmic spondylolysis and spondylolisthesis has never been fully documented. The recommendations for patients who need treatment and those who do not therefore have been based largely on studies of patients with symptoms or on a surgeon's personal opinion and experience, or both. We hope that the studies we report here will clarify the natural history and provide a solid basis for prognosis in a patient for whom active treatment is not undertaken.

It has been shown by others[4,8,11,24] that isthmic spondylolysis with or without spondylolisthesis does not exist at birth, and our limited study of roentgenograms confirms this observation. A possible exception was reported by Borkow and Kleiger[6]. They clinically diagnosed spondylolisthesis in a newborn; however, no roentgenograms were made until the patient was four months old. Subsequent surgery confirmed the presence of a bilateral pars interarticularis defect. Wiltse[39] and Wertzberger and Peterson[35] reported cases of patients with spondylolisthesis diagnosed at the age of eighteen months. Wertzberger and Peterson specifically stated that the defect developed without the patient having symptoms of pain or discomfort[35].

Our main study showed that the incidence of spondylolysis at the fifth lumbar vertebra was 4.4 per cent in six-year-old children and increased to 5.8 per cent in adulthood. These figures are similar to those in previously reported studies[3,7,8,13,17,19,23,26,28], although the reported

incidence in some selected populations has been much higher[31]. Our male:female ratio of nearly 2:1 is similar to that reported by Roche and Rowe[23].

Many authors have proposed various roentgenographic and clinical measurements and signs to predict whether a patient with spondylolysis will undergo progressive olisthesis or not. Taillard suggested that two anatomical factors play an important role in the development of a slip: the shape of the fifth lumbar vertebra and the shape of the dome of the sacrum[32]. He concluded that when the trapezoidal shape of the fifth lumbar vertebra was abnormal, with a lumbar index of less than 65 per cent associated with a rounded dome of the sacrum, more slippage was likely to occur. The lumbar vertebra in our control subjects showed an over-all gradual shift away from the trapezoidal shape that is usually present in childhood toward the normal rectangular shape present in adults. This was in contradistinction to the patients with a pars interarticularis defect, in whom the vertebra developed more of a trapezoidal contour. The patients who had an increase in olisthesis had a larger increase in the trapezoidal shape of the fifth lumbar vertebra than those who had no change. The male patients showed more of this change than did the female patients.

There was no evidence in our series that a change in the spondylolytic fifth lumbar vertebra to a more trapezoidal shape occurred before the observed increase in olisthesis. The change in the shape of the fifth lumbar vertebra occurred at the same time that the slip appeared or soon afterward. Thus, we found no correlation between our data and Taillard's two prognostic parameters as predictors of further slip. It is our opinion that these changes in the shape of the vertebrae are the result of the slip, rather than the cause.

In several of our subjects a pars interarticularis defect developed while they were under observation (Fig. 5). Two had a unilateral defect on initial examination and later had a bilateral defect, while in eight a bilateral defect developed in what appeared to be a normal vertebra. Additional unrecognized pars interarticularis defects probably exist in the children who were lost to follow-up from our initial 500. None of the children whom we saw with a defect complained of discomfort during the time that the defect developed.

In seven patients there was an increase in percentage of slip: in four male patients who had an established spondylolysis by the age of six and in three patients, who were found to have a pars interarticularis defect after the age of six, at about the same time that the pars interarticularis defect appeared. None of these children complained of pain during this period of time. In 1957, Schneider and Melamed reported the case of a child in whom a pars interarticularis defect developed, with a subsequent slip[25]. They unfortunately did not mention if the child had pain while under observation. A fusion was done about ten years later[25]. Taillard reported on a series of fifty patients with spondylolisthesis, all of whom had pain. Twelve had an unquestionable increase in slip before the age of twenty[32]. Turner and Bianco reported on a series of 173 children and teen-agers with spondylolisthesis[34]. One hundred and thirty-nine of them were seen for pain and the remainder, for deformity. Turner and Bianco did not observe progression of slip in any of the patients, all of whom were treated conservatively. Wiltse suggested that most slips occur between the ages of ten and fifteen, and that few are symptomatic[41]. Our study confirms the observations that progression of a slip can occur between the age of six and adulthood and that progression is not the rule. The olisthesis in any one patient seems to occur concomitantly with the development of the pars interarticularis defect. It was unusual, in our study, for progressive olisthesis to develop in a patient once the diagnosis

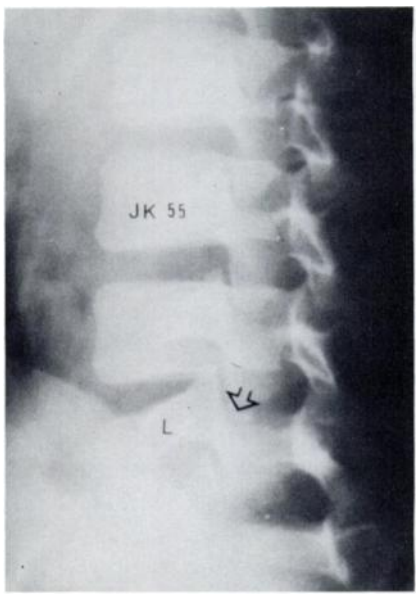

FIG. 5-A

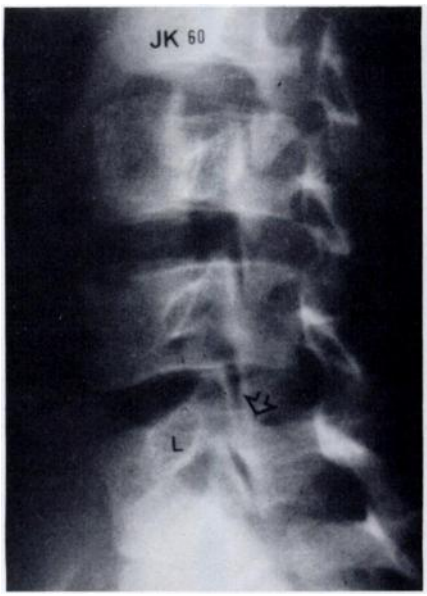

FIG. 5-B

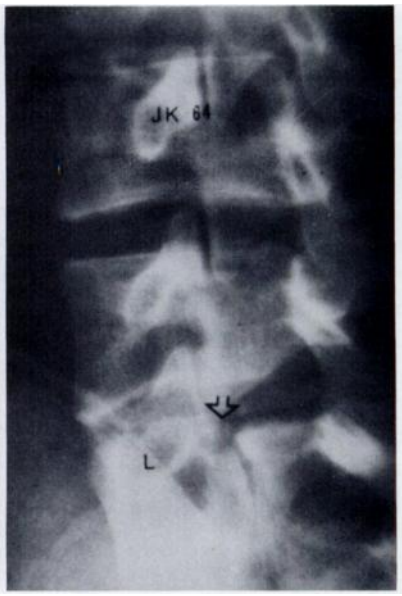

FIG. 5-C

Fig. 5-A: Roentgenogram of a six-year-old patient with no evidence of spondylolysis.
Fig. 5-B: Roentgenogram of the same patient at the age of eleven, with no evidence of spondylolysis.
Fig. 5-C: Roentgenogram of the patient at the age of fifteen, showing a bilateral defect at the fifth lumbar vertebra and a slip of 10 per cent.

of spondylolysis had been made. This was observed in only four male patients (Fig. 6). We were unable to ascertain the ages during which the children were particularly susceptible to progressive olisthesis. Progression was never noted after the age of sixteen. Our limited data on postpartum women suggest that in young women there is little chance of further slip with pregnancy. Our data cannot support the contention that neither the development of a pars interarticularis defect nor the progression of the slip tend to be painful. Our data are specifically restricted to isthmic spondylolisthesis in patients from the ages of six to twenty-five years.

It is common practice today to make serial roentgenograms for all children who are found to have evidence of spondylolysis or spondylolisthesis. If there is an increase in olisthesis, spine fusion is indicated, according to many authorities. This practice is based on data derived from observations of children who have had pain or significant deformity as well as alterations in gait and posture presumed to be secondary to the spondylolisthesis. We believe that it is incorrect to apply these principles to asymptomatic children with spondylolysis or isthmic spondylolisthesis, or both. These children, as was well documented in our study, usually do not have symptoms or further significant progressive olisthesis. We recommend, therefore, that the parents of the asymptomatic child with a pars interarticularis defect be advised of its existence, and be told to have the child re-evaluated only if symptoms develop. We do not restrict the child's activity in any way, and we allow full participation in sports.

Our data on sacral inclination, lumbar lordosis, and percentage of slip were obtained from roentgenograms made with the patient in the recumbent position. We concur with previous authors[15] who have documented that there is a significant difference in these measurements when the roentgenograms are made with the patient in the erect compared with the recumbent position. We think, therefore, that our data on lumbar lordosis and sacral inclination cannot be compared with previously published data on patients who were in an erect position when the roentgenogram was made. We also recognize that our data on percentages of slip might have been different if the roentgenograms had been made with the patient in that position.

Architectural abnormalities such as lumbarization, sacralization, and spina bifida occulta have been associated with spondylolysis to varying degrees. Wiltse stated that spina bifida occulta is seen thirteen times more often in patients with spondylolysis than in those without that lesion[42]. Taillard reported a 42 per cent incidence of spina bifida occulta in patients with spondylolysis[32]; Laurent and Einola, 22 per cent[14]; Friberg, 28 per cent[8]; and Meyerding, 35 per cent[16]. In the study by Laurent and Einola, lumbarization or sacralization was present in 9 per cent of the patients with spondylolysis[14]. The incidence of spina bifida occulta in our series varied with the ages of the group studied: it was 59 per cent in the six-year-old children without spondylolysis and 20 per cent in the adults. These figures are similar to those reported in the literature. It should be noted, however, that spina bifida occulta rarely occurs in the lumbar segments of normal adults. In most of the adults in our study with spina bifida occulta only the sacrum was involved, while the lumbar segments were involved in only 7 per cent.

Spina bifida occulta was found in 92 per cent of the children with a pars interarticularis defect; 55 per cent of these children had a defect in the lumbar region in addition to a defect in the sacrum. In 70 per cent of the children the defect persisted into adulthood, and in 30 per cent the defect was inclusive of the fifth lumbar vertebra. This association

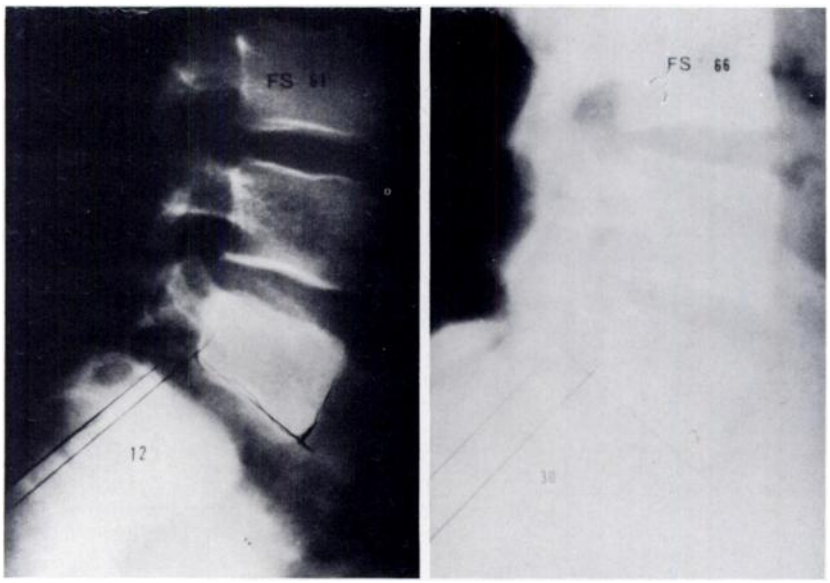

FIG. 6-A FIG. 6-B

Figs. 6-A and 6-B: Roentgenograms of a patient who was six years old when a bilateral defect at the fifth lumbar vertebra was detected.
Fig. 6-A: The patient at the age of eleven, showing a 12 per cent slip.
Fig. 6-B: The patient at the age of sixteen, showing an increase in the slip to 30 per cent.

of spina bifida occulta and spondylolysis has been noted in the past[42]. It certainly supports the argument that the defect has an inheritance factor. The incidence of lumbarization and sacralization in the group without a pars interarticularis defect was 16 per cent, and in those with a defect the incidence was 7 per cent. This difference is not statistically significant.

It is generally agreed that inheritance plays a major role in the development of the pars interarticularis defect[38,40-42,44], and the data from our first ancillary study supported this idea. Wiltse suggested that there is a hereditary defect or dysplasia in the cartilaginous model of the arch of the affected vertebra, and that there is a recessive pattern of inheritance with varying expressivity that is not sex-linked[40]. In a later paper, he suggested the possibility of incomplete dominance of a gene in certain patients[42]. Wynne-Davies and Scott, in 1979, reported a study of forty-seven families, thirty-five of which had isthmic spondylolysis[45]. All of the probands were obtained through an orthopaedic clinic. Fifteen per cent of the relatives were similarly affected. Wynne-Davies and Scott concluded that the pattern of inheritance was either autosomal dominant with reduced penetrance or was multifactorial. Our data alone, and our data combined with pedigrees from the literature, do not allow us to discriminate between the two hypotheses: (1) genetic heterogeneity with multiple mendelian forms, or (2) multifactorial inheritance with some family members having a higher liability than others. In order to resolve the question, more extended family studies must be undertaken. We do not believe that it is possible, in the United States today, to identify the large groups of asymptomatic families that would be needed for such a study.

Environmental factors may be an important ancillary factor in the development of a pars interarticularis defect. This study was carried out during a time when girls were not as active in sports as boys, which might have made a defect less likely to develop in the girls even though they had similar genetic susceptibility. This factor, however, may not be as relevant as some consider it to be, since up to the age of five years the activity levels of boys and girls are similar, and the ratio of boys to girls was the same (2:1) as the sex ratio in the adults in our series. There was no evidence (pain) to suggest that the patients with spondylolysis or spondylolisthesis found after the age of six had a non-united stress fracture.

Conclusions

Based on the results of our study and on the data in the literature, the following conclusions can be drawn. The incidence of spondylolysis, with or without spondylolisthesis, is 4.4 per cent at the age of six compared with about 6 per cent in adulthood. The male:female ratio of affected subjects is 2:1 at the age of six and in adulthood. Spondylolisthesis does not exist at birth.

Slipping, when it occurs, usually is demonstrable at about the same time that the pars interarticularis defect is first detected roentgenographically. Slipping may increase up to the age of sixteen, but does so rarely. Development of the pars interarticularis defect, with or without spondylolisthesis, does not cause pain in most patients. A trapezoidal shape of the fifth lumbar vertebra is probably a result of the slipping, not a cause. Spina bifida occulta occurs more frequently in patients with a pars interarticularis defect than in patients without a defect.

Inheritance is a factor in the development of a pars interarticularis defect, probably based on either genetic heterogeneity with multiple mendelian forms or on multifactorial inheritance, with some family members having a higher liability to the lesion than others.

We suggest that a child with spondylolysis or spondylolisthesis can be permitted to enjoy a normal childhood and adolescence without restriction of activities and without fear of progressive olisthesis or disabling pain.

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References

1. AMERICAN ACADEMY OF ORTHOPAEDIC SURGEONS, COMMITTEE ON THE SPINE: A Glossary on Spinal Terminology. Chicago, The American Academy of Orthopaedic Surgeons (no date).
2. AMUSO, S. J., and MANKIN, H. J.: Hereditary Spondylolisthesis and Spina Bifida. J. Bone and Joint Surg., **49-A:** 507-513, April 1967.
3. BAILEY, WILBUR: Observations on the Etiology and Frequency of Spondylolisthesis and Its Precursors. Radiology, **48:** 107-112, 1947.
4. BATTS, MARTIN, JR.: The Etiology of Spondylolisthesis. J. Bone and Joint Surg., **21:** 879-884, Oct. 1939.
5. BLACKBURNE, J. S., and VELIKAS, E. P.: Spondylolisthesis in Children and Adolescents. J. Bone and Joint Surg., **59-B(4):** 490-494, 1977.
6. BORKOW, S. E., and KLEIGER, BARNARD: Spondylolisthesis in the Newborn. A Case Report. Clin. Orthop., **81:** 73-76, 1971.
7. CONGDON, R. T.: Spondylolisthesis and Vertebral Anomalies in Skeletons of American Aborigines. With Clinical Notes on Spondylolisthesis. J. Bone and Joint Surg., **14:** 511-524, July 1932.
8. FRIBERG, STEN: Studies on Spondylolisthesis. Acta Chir. Scandinavica, Supplementum 55, 1939.
9. GEORGE, E. M.: Spondylolisthesis. Surg., Gynec. and Obstet., **68:** 774-781, 1939.
10. HAUKIPURO, K.; KERANEN, N.; KOIVISTO, E.; LINDHOLM, R.; NORIO, R.; and PUNTO, L.: Familial Occurrence of Lumbar Spondylolysis and Spondylolisthesis. Clin. Genet., **13:** 471-476, 1978.
11. HITCHCOCK, H. H.: Spondylolisthesis. Observations on Its Development, Progression, and Genesis. J. Bone and Joint Surg., **22:** 1-16, Jan. 1940.
12. HOSHINA, HITOSHI: Spondylolysis in Athletes. Phys. and Sportsmed., **8:** 75-79, Sept. 1980.
13. LANIER, R. R., JR.: The Presacral Vertebrae of American White and Negro Males. Am. J. Phys. Anthropol., **25:** 341-420, 1939.
14. LAURENT, L. E., and EINOLA, S.: Spondylolisthesis in Children and Adolescents. Acta Orthop. Scandinavica, **31:** 45-64, 1961.
15. LOWE, R. W.; HAYES, T. D.; KAYE, JEREMY; BAGG, R. J.; and LUEKENS, C. A., JR.: Standing Roentgenograms in Spondylolisthesis. Clin. Orthop., **117:** 80-84, 1976.
16. MEYERDING, H. W.: Spondylolisthesis. J. Bone and Joint Surg., **13:** 39-48, Jan. 1931.
17. MORETON, R. D.: Spondylolysis. J. Am. Med. Assn., **195:** 671-674, 1966.
18. MORTON, N. E.: Segregation and Linkage. *In* Methodology in Human Genetics, pp. 17-52. Edited by W. J. Burdette. San Francisco, Holden-Day, 1962.
19. NEUGEBAUER, F. L.: The Classic: A New Contribution to the History and Etiology of Spondyl-olisthesis. Clin. Orthop., **117:** 4-22, 1976.
20. NEWMAN, P. H., and STONE, K. H.: The Etiology of Spondylolisthesis. J. Bone and Joint Surg., **45-B(1):** 39-59, 1963.

21. OHTA, H.: Spondylolysis: Familial Occurrence and Its Genetic Implication. J. Japanese Orthop. Assn., **41:** 931-941, 1967.
22. PERRET, GEORGE: Congenital and Developmental Anomalies. Skeletal and Clinical Manifestations of Anomalies and Defects of the Neuraxis. Clin. Orthop., **27:** 9-28, 1963.
23. ROCHE, M. B., and ROWE, G. G.: The Incidence of Separate Neural Arch and Coincident Bone Variations. A Survey of 4,200 Skeletons. Anat. Rec., **109:** 233-252, 1951.
24. ROWE, G. G., and ROCHE, M. B.: The Etiology of Separate Neural Arch. J. Bone and Joint Surg., **35-A:** 102-110, Jan. 1953.
25. SCHNEIDER, C. C., and MELAMED, ABRAHAM: Spondylolysis and Spondylolisthesis. Case Report Clarifying the Etiology of Spondylolysis. Radiology, **69:** 863-866, 1957.
26. SHAHRIAREE, H.; SAJADI, K.; ROOHOLAMINI, S. A.; and SHEIKH, S.: A Family with Spondylolisthesis. Scientific Exhibit at the Annual Meeting of The American Academy of Orthopaedic Surgeons, Las Vegas, Nevada, Feb. 3-8, 1977.
27. SHANDS, A. R., JR., and BUNDENS, W. D.: Congenital Deformities of the Spine. An Analysis of the Roentgenograms of 700 Children. Bull. Hosp. Joint Dis., **17:** 110-133, 1956.
28. SHORE, L. R.: Abnormalities of the Vertebral Column in a Series of Skeletons of Bantu Natives of South Africa. J. Anat., **64:** 206-238, 1930.
29. SOREN, ARNOLD: Spondylolisthesis and Trauma. Indust. Med. and Surg., **34:** 917-921, 1965.
30. SPLITHOFF, C. A.: Lumbosacral Junction. Roentgenographic Comparison of Patients with and without Backaches. J. Am. Med. Assn., **152:** 1610-1613, 1953.
31. STEWART, T. D.: Incidence of Separate Neural Arch in the Lumbar Vertebrae of Eskimos. Am. J. Phys. Anthropol., **16:** 51-62, 1931.
32. TAILLARD, W.: Le spondylolisthesis chez l'enfant et l'adolescent (Etude de 50 cas). Acta Orthop. Scandinavica, **24:** 115-144, 1954.
33. TOLAND, J. J., III: Spondylolisthesis in Identical Twins. Clin. Orthop., **5:** 184-189, 1955.
34. TURNER, R. H., and BIANCO, A. J., JR.: Spondylolysis and Spondylolisthesis in Children and Teen-Agers. J. Bone and Joint Surg., **53-A:** 1298-1306, Oct. 1971.
35. WERTZBERGER, K. L., and PETERSON, H. A.: Acquired Spondylolysis and Spondylolisthesis in the Young Child. Spine, **5:** 437-442, 1980.
36. WILLIS, T. A.: The Lumbo-Sacral Vertebral Column in Man, Its Stability of Form and Function. Am. J. Anat., **32:** 95-123, 1923.
37. WILLIS, T. A.: The Separate Neural Arch. J. Bone and Joint Surg., **13:** 709-721, Oct. 1931.
38. WILTSE, L. L.: Spondylolisthesis. Personal communication.
39. WILTSE, L. L.: Spondylolisthesis. Etiology and Comparative Efficacy to Various Procedures. Personal communication.
40. WILTSE, L. L.: Etiology of Spondylolisthesis. Clin. Orthop., **10:** 48-59, 1957.
41. WILTSE, L. L.: Spondylolisthesis in Children. Clin. Orthop., **21:** 156-163, 1961.
42. WILTSE, L. L.: The Etiology of Spondylolisthesis. J. Bone and Joint Surg., **44-A:** 539-560, April 1962.
43. WILTSE, L. L., and HUTCHINSON, R. H.: Surgical Treatment of Spondylolisthesis. Clin. Orthop., **35:** 116-135, 1964.
44. WILTSE, L. L.; WIDELL, E. H., JR.; and JACKSON, D. W.: Fatigue Fracture: The Basic Lesion in Isthmic Spondylolisthesis. J. Bone and Joint Surg., **57-A:** 17-22, Jan. 1975.
45. WYNNE-DAVIES, RUTH, and SCOTT, J. H. S.: Inheritance and Spondylolisthesis. A Radiographic Family Survey. J. Bone and Joint Surg., **61-B(3):** 301-305, 1979.