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Prevalence of Sacral Spina Bifida Occulta and Its Relationship to Age, Sex, Race, and the Sacral Table Angle

An Anatomic, Osteologic Study of Three Thousand One Hundred Specimens

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Study Design. An anatomic, osteologic study of spina bifida occulta (SBO).

Objective. To determine the prevalence and patterns of SBO in a large population and examine its relationship to age, sex, and race; then to evaluate SBOs relationship to the sacral table angle (STA) when compared with an age-matched control group.

Summary of Background Data. SBO has a reported prevalence of 1.2% to 50% and has been implicated in various pathologic problems. SBO is often associated with spondylolysis or spondylolithesis. The STA has been implicated as an etiologic or predictive factor in the presence of pars defects.

Methods. Three thousand one hundred osteologic specimens were evaluated for the presence of SBO. SBO was graded on a scale from 0 to III. Information on the age, sex, race, and STA of each specimen was recorded and measured, respectively. Prevalence and patterns of SBO were enumerated. The STAs of an age-matched control group of 355 specimens were examined. The SBO group and control groups were compared in regards to STA, controlling for age, sex, and race.

Results. Overall, 355 specimens displayed SBO, for an overall prevalence of 12.4%. Of the SBO specimens, 68.7% were white, 88.2% were men, 53% were grade I, 37% II, and 10% III. All 3 grades of SBO were more common in men than women (88.2% vs. 11.8%) and more prevalent in whites than blacks (68.7% *vs.* 31.3%) ($P = 0.01$). SBO decreased in prevalence with increasing age. The average STA in SBO specimens was 95.9°. This differed from an age-matched control group, 92.1° ($P < 0.0001$). Every 1° increase in STA resulted in a 6% increased likelihood of SBO. In SBO specimens, the STA decreased with increasing age, contrary to age-matched controls.

Conclusion. SBO has an overall prevalence of 12.4% in a large, diverse population. SBO is more common in men and whites and decreases in prevalence with increasing age. The STA is greater in SBO when compared with controls and an increased STA predicts SBO. In SBO, the STA decreases with increasing age.

Key words: spina bifida occulta, sacral table angle, prevalence, spondylolisthesis. **Spine 2009;34:1539–1543**

Spina bifida occulta (SBO) is on the spectrum of spinal dysraphisms, representing an incomplete closure in the posterior elements of the neural arch.[1,2] The prevalence of SBO has been variously reported, ranging from 1.2% to 50% in different study populations.[2,3] Much of the reported variation can be attributed to the different distributions, or lack thereof, in age, gender, and race in the populations studied.[2] A large, radiographic study by Fidas *et al*[2] determined an overall prevalence of 23% in a racially uniform, Scottish population.

Despite the frequent appearance of SBO in clinical practice, controversy exists as to its importance. Although generally considered a minor radiographic abnormality, SBO has been associated with a panoply of problems ranging from urodynamic difficulties[2,4] to posterior disc herniations[3] and increased low back pain.[5] SBO often accompanies spondylolysis and spondylolisthesis[6]; the 2 entities appear collectively in 33% to 61% of spondylolysis or isthmic spondylolisthesis cases.[7,8] However, no consensus exits as to whether the presence of SBO affects the stability of the lumbosacral junction, particularly when associated with a spondylolytic defect.[8,9] Recent attention to mechanical stability in spondylolisthesis has focused on the sacral table angle (STA) as an associated and possible etiologic factor.[10,11] It has been suggested that patients with a lower STA are predisposed to progressive slip.[12] Given the strong connection between SBO and spondylolisthesis, it seems to follow that the STA should be similarly decreased in SBO when compared with controls.

The present study, therefore, has 2 aims. First, to definitively define the prevalence of SBO through an anatomic study of a racially diverse population, the largest study of SBO yet reported in the literature; and then, to define the patterns of SBO in relation to age, sex, and race. Second, to examine the relationship between SBO and STA, with the hypothesis being that STA is decreased in SBO when compared with normal controls.

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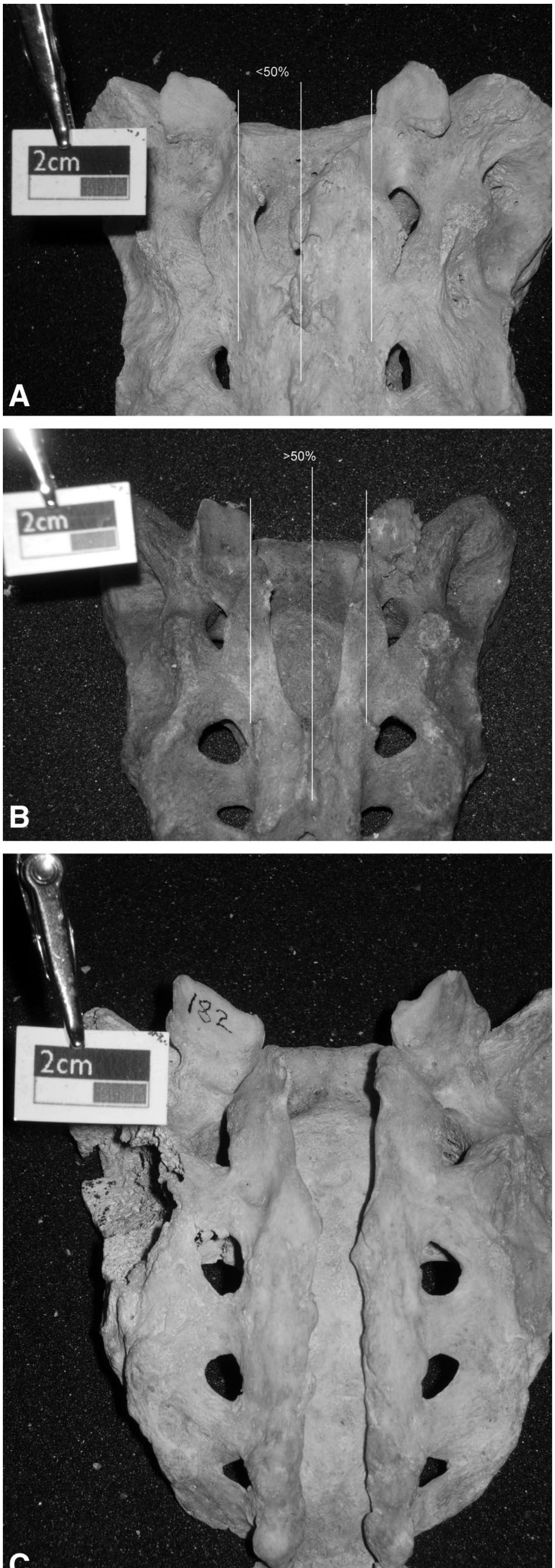

Figure 1. **A,** Partial SBO defect (<50%). **B,** Complete SBO defect (>50%). **C,** Pan-sacral SBO defect (opening: S1–S5).

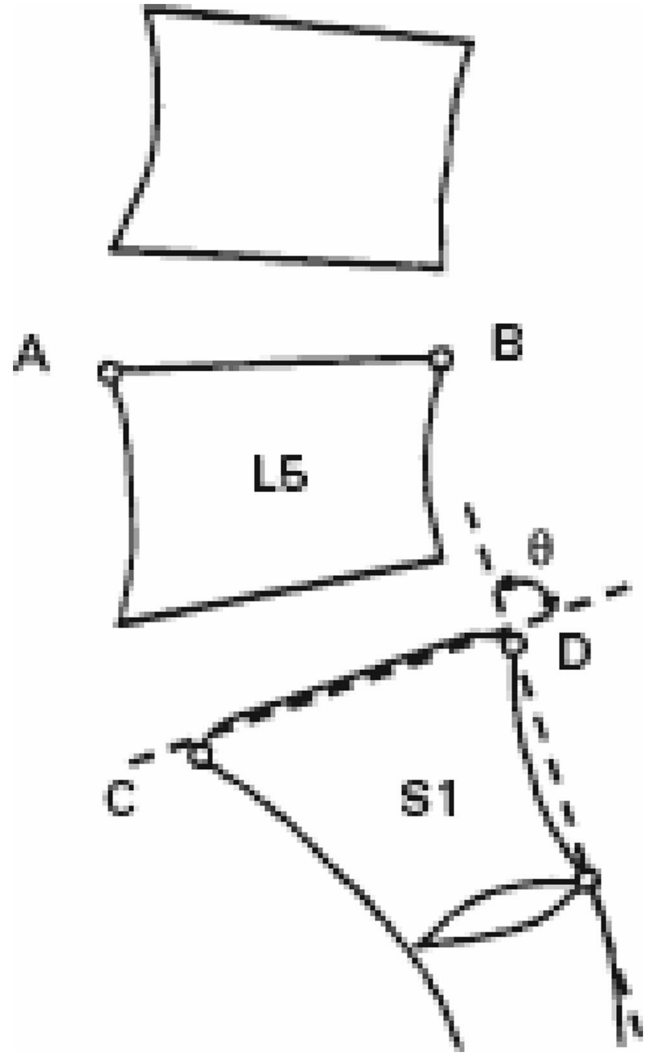

Figure 2. STA represents the angle between a line drawn along the posterior aspect of S1 and a line drawn along the sacral end plate (Reprinted with permission from Spine 2002;27:831–8).

■ Materials and Methods

This study of SBO was an osteologic, anatomic examination of the 3100 human specimens representing the Hamann-Todd Osteological Collection in Cleveland, Ohio. This collection represents skeletal remains collected between 1893 and 1938 from individuals living in the Cleveland, Ohio area. Of the total 3100 specimens, 234 specimens were excluded because of lost/damaged sacrums or L5 sacralization/S1 lumbarization, leaving 2866 specimens available for final review. This represents the largest population study of SBO reported in the literature. SBO was graded according to the percentage of opening in the

Table 1. Specimens by Age Category

Age Categories	SBO	Controls
<30	75	62
30–39	62	57
40–49	92	94
50–59	65	72
60–69	45	49
>70	21	25

Table 2. Prevalence of SBO by Decade

Age (Total in Category)	% SBO
<30 (n = 384)	19.5%
30–39 (n = 520)	11.9%
40–49 (n = 677)	13.6%
50–59 (n = 610)	10.7%
60–69 (n = 442)	10.2%
>70 (n = 309)	6.8%

posterior neural arch. This grading system, modeled on the recommendations of Kettler and Wilke,[13] was developed to categorize the SBO defects on a scale from 0 to III. Beginning with a normal state, grade 0 indicated an unaffected, complete posterior arch. Grade I ("partial") was defined as a defect with <50% opening in the posterior arch of S1–S2. Grade II ("complete") was defined as >50% opening of the S1–S2 arch. And grade III ("pan-sacral") was defined as failure of closure from S1–S5, irrespective of percentage of opening (Figures 1A–C). When applicable, the same grading system (grades: 0–II) was used to evaluate SBO lesions in the lumbar spine.

Those specimens (n = 355) demonstrating evidence of SBO with this system represented group I. To fulfill the second aim of the study (*i.e.*, analysis and comparison of STA), a group II was selected from unaffected, age-matched control specimens (n = 355). This control group was randomly selected from remaining unaffected individuals (n = 2511) in the original study group (n = 2866). These controls were randomly pulled from the shelves according to age and ease of access in the collection. The STA represents the angle between a line drawn along the sacral end plate and a line drawn along the posterior aspect of the S1 vertebral body (Figure 2).[10] The sacral end plate of S1 was defined by its relationship to the sacral ala. Normally, the superior end plate of S1 sits below the prominence of the sacral ala. However, if the end plate was obviously higher than the sacral ala, this was assumed to be a sacralized L5 vertebral end plate and the specimen was excluded as previously mentioned. The STA was measured with a protractor method as described by Whitesides *et al.*[11] This method has been previously shown to produce an error of 1.1° when compared with radiographic measurement.[11] All specimens were examined and measured by the primary author (J.D.E.). An intraclass correlation test was done to determine the intraobserver reliability of these measurements. Fifty specimens were measured by a single examiner on 3 separate occasions and the ICC was 0.995 with a 95% confidence interval (0.997, 0.999), indicating excellent intraobserver reliability.

To assess the significance of difference in STA measurements between SBO and control subjects, a power analysis was performed. Whitesides *et al*[11] previously demonstrated differences in STA values of approximately 3° to 5° between spondylolytic and normal specimens. Using this as a model, with a power of 95% and a $P < 0.05$ (difference in means of 3 and a standard deviation of 7), it was determined that 143 specimens would be required to show a significant result. With 355 specimens, the present study had ample power to detect important differences in STA.

Table 3. Specimens by Race

Race	SBO	Controls
African American	110	155
White	245	199
American Indian	0	1

Table 4. Specimens by Gender

Gender	SBO	Controls
Women	42	67
Men	313	288

Statistical analysis was then performed to evaluate for any significant differences in the groups. ANOVAs were employed to assess differences in SBO grade by age and STA, followed by multiple comparison tests using Bonferroni adjustment factor. χ^2 tests were used for independence of attributes to test association of race and gender with SBO grade. Ordinal logistic regression was used to assess if STA predicts grade of SBO, while 2 sample *t* tests were conducted to assess for a significant difference in STA between specimens and controls. Finally, a binary logistic regression model was employed to study if STA predicts SBO when controlled for age, sex, and race.

■ Results

Of the 2866 specimens available for final analysis, 355 specimens were noted to demonstrate SBO defects, for an overall prevalence of 12.4%. When broken down into categories, SBO was identified in 8.3% women, 13.2% men, 14.2% white, and 9.8% of the black specimens. Tables 1 and 2 delineate the total number of SBO specimens by age (compared with controls) and the decade specific prevalence, respectively. Tables 3 and 4 demonstrate the categorization by race and gender, respectively. SBO was twice as common in men as women and 46% more common in whites than blacks. As has been previous demonstrated,[2] the prevalence of SBO appeared to decrease with increasing age (Figure 3).

In regards to grade, 53% of the SBO specimens displayed grade I defects, 37% grade II, and 9.8% grade III (Figure 4). Mean age of grade I (42.3 years) was less than grade II (48.1) ($P = 0.006$); however, no difference was found between grades I/III and grades II/III ($P = 0.38$ and $P = 1$). All 3 grades were more common in whites ($P = 0.01$) and men ($P = 0.01$). There was no statistically

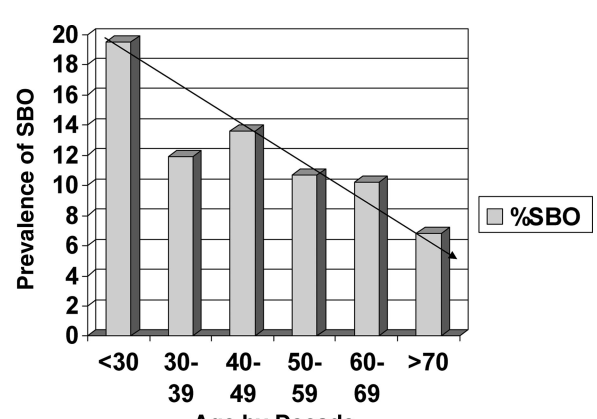

Figure 3. Prevalence of SBO by decade. Note here the gradual decrease in prevalence with increasing age.

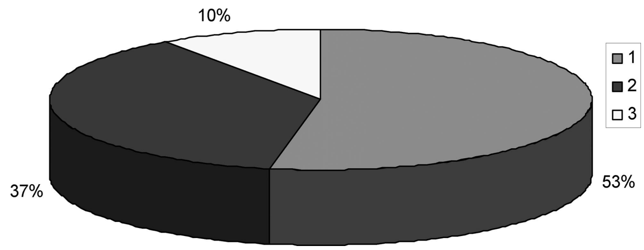

Figure 4. Breakdown of SBO specimens by grade of defect.

significant association between grade of SBO and STA ($P = 0.25$).

The average STA in SBO specimens was 95.9° ± 7.0°, while the average in controls was 92.1° ± 6.6° ($P < 0.001$). The average STA in SBO specimens decreased linearly with increasing age (Figure 5A), whereas no such definitive pattern was noted in age-matched control specimens (Figure 5B). Compared with controls, every 1° increase in STA resulted in a 6% increased likelihood of SBO.

In the SBO population studied, there were 20 specimens with concomitant grade I L5 occulta and 1 specimen with grade I L4 occulta. There were 38 specimens with L5 spondylolysis (~11%) in the SBO group. This represents an increased number compared with non-SBO subjects, a finding consistent with previous reports.[8] Eight of these L5 spondylolytic specimens were associated with grade III occulta (~21%). The average STA in SBO and spondylolytic specimens was 93.5°.

Discussion

SBO has a high prevalence in the axial skeleton. As the largest and most racially diverse study in the literature, this study found an overall prevalence of 12.4%, in keeping with the reported range of 1.2% to 50%.[2] As has been previously shown, analysis of the specimens showed a higher prevalence of SBO in men than in women.[2] Additionally, this study showed a higher prevalence of SBO in whites than blacks, indicating probable genetic and racial differences.[2] Apparently, SBO is somewhat of an age dependant, "disappearing" entity. In other words, this study showed a decreasing prevalence of SBO with increasing age, confirming the findings of Fidas *et al*[2] who postulated that this represents increasing ossification of the defects with age associated degenerative disease. Although the present study demonstrated various forms of SBO, ranging from mild defects to pansacral openings, no significant correlation could be found between grade of SBO and age, sex, or race. This raises the question then as to the clinical importance of SBO defects.

Although the tendency is to consider SBO defects benign given their high prevalence, numerous authors[2,3,8,14] have warned against making this conclusion. SBO has been implicated in urodynamic irregularities[2,4] and hip dysplasia.[15] It has been associated with increased numbers of posterior disc herniations[3] and low back pain.[5] Other authors[8,14] warn that SBO, when not carefully considered, can lead to intraoperative neurologic injury. Finally, the strong association between SBO and spondyloysis/spondylolisthesis has led some to suggest a possible mechanical difference in the lumbosacral junction of SBO patients.[8,16]

Concerning the anatomic and mechanical forces at the lumbosacral junction with potential contribution to spondylolisthesis, recent work has associated a predisposition to further slip with the STA.[12] Further, Whitesides

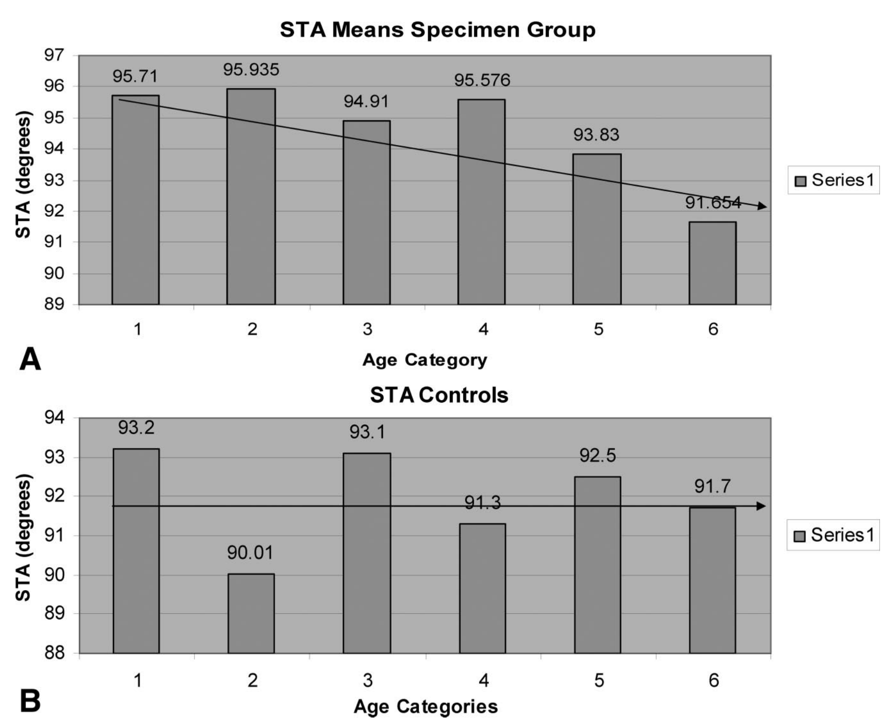

Figure 5. **A,** STA in SBO individuals. Note here the gradual decrease in STA with increasing age. **B,** STA in controls. In comparison with the SBO specimens, note here the lack of a true downward trend in STA angle.

et al[11] suggests that the STA may be etiologic in the development of spondylolytic spondylolisthesis. There is a documented high association between spondylolysis/spondylolisthesis and SBO.[6–8] Understanding this association and correlating it with the STA findings of Hosoe and Ohmori[12] concerning degenerative, lumbosacral spondylolisthesis, the present study hypothesized that SBO would be accompanied with a lower STA. Curiously, this study showed just the opposite. SBO specimens had significantly higher STAs when compared to age-matched controls. In fact, the STA was predictive of SBO, with every 1° increase in STA increasing the likelihood of SBO by 6%. Perhaps as a kind of compensation for the absence of posterior stabilizing attachments, the sacral table assumes a more horizontal position in the presence of SBO. This developmental attempt at increasing stability at the lumbosacral junction may play less of a role as the spine ages. In this study, decreasing STAs were noted with increasing age in the SBO group, a trend which paralleled the overall decreased incidence of SBO in the population over time. Apparently, the sacral table remodels over time, becoming less horizontal, while at the same time the prevalence of SBO decreases. With increasing age, ossification of the posterior elements and progressive degenerative disease and disc collapse may add increasing stiffness/stability to the lumbosacral junction and lead to the development of a compensatory decrease in STA with time. Finally, the present study did not find any significant association between the grade of SBO and the STA, making it difficult to draw any definitive assumptions concerning correlation between the amount of posterior defect and any potential instability.

The limitations of this study are inherent in the study design. More specifically, this is an anatomic study performed on dried, disarticulated, osteologic specimens sitting in collection boxes. As such, this study neither takes into account the soft tissue or cartilaginous components at the lumbosacral junction, nor the natural, anatomic alignment of the individuals in the standing position. Similarly, this study does not include an examination of the L5–S1 facet tropism, which may affect the biomechanics of the SBO/STA relationship; this presents an interesting topic for future examination. Finally, the study is limited by the composition of the Hamann-Todd collection; the potential for bias between age groups does exist, as older specimens may represent healthy individuals who died of old age and younger specimens may represent unhealthy or traumatic deaths. Recognizing this possibility, however, the authors do not think such bias would affect a common anatomic finding like SBO. These limitations aside, the present study definitely establishes an association between an increased STA and the presence of SBO.

■ Conclusion

SBO is a common entity occurring in 12.4% of the population. It is more common in men than women and in whites than blacks. Partial defects are more common than complete ones. The prevalence of SBO decreases with increasing age. The STA is greater in SBO subjects than in age-matched controls and is predictive of the presence of SBO.

■ Key Points

- SBO is a common finding in the sacrum, occurring in approximately 12% of the population.
- SBO is more common in whites than blacks and men than women.
- Partial defects in the posterior arch of S1 and S2 are most common, followed by complete defects and pan-sacral defects (S1–S5).
- The STA is greater in SBO than normal controls.
- The STA is predictive of the presence of SBO.

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