



# Posterior Wall Defect of Sacrum: An Anatomical Study of Sacral Spina Bifida

Goksin Nilufer DEMIRCI YONGUC<sup>1</sup>, Salih SAYHAN<sup>1</sup>, Sibel CIRPAN<sup>1</sup>, Burcu BULUT<sup>2</sup>, Mustafa GUVENCER<sup>3</sup>, Sait NADERI<sup>4</sup>

<sup>1</sup>Dokuz Eylul University, School of Medicine, Department of Anatomy, Izmir, Turkey

<sup>2</sup>Hatay Mustafa Kemal University, School of Medicine, Department of Anatomy, Hatay, Turkey

<sup>3</sup>Izmir Tinaztepe University, School of Medicine, Department of Anatomy, Izmir, Turkey

<sup>4</sup>Health Sciences University, Umraniye Education and Research Hospital, Department of Neurosurgery, Istanbul, Turkey

This study has been presented as an oral presentation at the 30<sup>th</sup> Annual Scientific Congress of the Turkish Neurosurgical Society between 8 and 12 April 2016 at Antalya, Turkey.

This study has been presented as a poster presentation at the 17<sup>th</sup> TAKAD National Anatomy Congress of the Turkish Society of Anatomy and Clinical Anatomy between 5 and 9 September 2016 at Eskişehir, Turkey

Corresponding author: Mustafa GUVENCER ✉ mustafa.guvencer@gmail.com

## ABSTRACT

**AIM:** To investigate the incidence, types, morphological and morphometric properties of spina bifida on dry sacral bones.

**MATERIAL and METHODS:** 110 dry adult sacrum gathered from the bone collections of the Laboratory of the Anatomy Department of Dokuz Eylul University School of Medicine were examined. The parameters analysed were: 1) results of parameters related to the posterior sacral wall; 2) classification and rate of the closure defects; 3) classification of the top sacral vertebrae according to the shape of its superior surface; 4) presence of sacralisation and lumbalisation among sacrum with dorsal wall defects; 5) vertebral levels of apex of the sacral hiatus; and 6) vertebral levels of closure defects of the sacrum.

**RESULTS:** We determined 22/110 (20%) sacrum demonstrated spina bifida. Of these 22 sacrum, 4 (18.18%) showed complete and 18 (81.82%) showed incomplete spina bifida. We noted the coexistence of spina bifida with sacralisation (6/22 [27.27%]) and lumbalisation (5/22 [22.73%]). The types of defects were described and grouped as 'V' (Type 1), inverse 'V' (Type 2), window (Type 3), foramen (hole) (Type 4), sand watch (Type 5), narrow linear (Type 6), wide linear (Type 7), and bridged (Type 8). The shape of upper surfaces of the sacrum with spina bifida was grouped as: cavity (20/22, 90.9%), hump (1/22, 4.5%), and flat (1/22, 4.5%).

**CONCLUSION:** A precise definition of the anatomical variations of sacrum is essential for surgeons, particularly when operating using endoscopic techniques and for anaesthesiologists applying caudal epidural block.

**KEYWORDS:** Defect, Sacral hiatus, Sacrum, Spina bifida

## INTRODUCTION

The worldwide incidence of spina bifida has declined in recent years. However, a larger proportion of new patients present with more distal lesions (i.e. sacral

(2,10,35). The incomplete closure of the sacral canal is known as spina bifida occulta (SBO) and is comprise of anomalies ranging from the partial defect of the posterior arch of the sacral vertebrae top to sacral S1-S5 spina bifida (16).

Goksin Nilufer DEMIRCI YONGUC : 0000-0002-6300-5365  
Salih SAYHAN : 0000-0003-3594-8937  
Sibel CIRPAN : 0000-0002-0654-9139

Burcu BULUT : 0000-0002-5868-4498  
Mustafa GUVENCER : 0000-0001-6953-2898  
Sait NADERI : 0000-0002-6784-4270

**Table I:** The Results of Parameters Related to Posterior Sacral Wall

|   | n  | Minimum-Maximum (mm) | Mean ± SD (mm) |
|---|----|----------------------|----------------|
| Horizontal diameter of the sacral canal superiorly (A)                              | 22 | 22.68-34.33          | 29.50 ± 3.51   |
| Anteroposterior diameter of the sacral canal superiorly (B)                         | 20 | 7.97-22.28           | 14.59 ± 4.47   |
| posterior height of the sacrum (C)  | 21 | 97.23-137.13         | 113.00 ± 11.21 |
| The distance between the median sacral crest and the upper margin of the sacrum (D) | 18 | 19.51-48.75          | 32.19 ± 9.53   |
| The length of the median sacral crest (E)   | 18 | 27.31-93.14          | 61.04 ± 20.24  |
| The height of apex of the sacral hiatus (F)   | 20 | 3.54-47.12           | 24.51 ± 11.30  |
| The distance between sacral corns at sacral hiatus (G)                              | 22 | 10.54-22.22          | 16.47 ± 2.90   |
| The A-P distance at apex of sacral hiatus (H)                                       | 20 | 1.37-11.89           | 5.63 ± 2.51    |

**Table II:** The Classification and the Rate of Posterior Sacral Wall Closure Defects

|            | Shape                       | Case (Rate) |
|------------|-----------------------------|-------------|
| Incomplete | “V” shaped (Type 1)         | 4 (18.18%)  |
|            | Inverse “V” shaped (Type 2) | 4 (18.18%)  |
|            | Window shaped (Type 3)      | 4 (18.18%)  |
|            | Foramen shaped (Type 4)     | 2 (9.09%)   |
|            | Sand watch shaped (Type 5)  | 0 (0%)      |
|            | Type 3+Type 5               | 1 (4.55%)   |
|            | Narrow linear (Type 6)      | 1 (4.55%)   |
|            | Type 1+Type 4               | 1 (4.55%)   |
|            | Type 3+Type 6               | 1 (4.55%)   |
|            | Wide linear (Type 7)        | 1 (4.55%)   |
| Complete   | Bridged (Type 8)            | 1 (4.55%)   |
|            | Type 5+Type 6               | 1 (4.55%)   |
|            | Type 1+Type 2+Type 3+Type 6 | 1 (4.55%)   |

The rate of sacral SBO (SSBO) alone has been observed in healthy populations as 12.4% to 21.4% (16,33). Degenerative calcification may cause the filling of some bone defects in the elderly population. Therefore, the prevalence of SBO tends to decrease with age (8,19). This age-related decrease in the prevalence of SBO has been documented many studies (8, 19,48,65,69).

It has been determined that 90% to 100% of cases with sacral level lesions are outpatients (1,9,20,30,58). The clinical spectrum of SBO ranges from insignificant cases with minimal anatomical variation to more severe cases of neurological deficits with meningocele (1,9,31,44,46,51,58,59,73). The skin and nervous system malformations may coincidentally be observed together owing to their common ectodermic origin (28). Sacral skin lesions such as dimples,

focal hypertrichosis, nevus, or lipomas are observed in 51% to 100% of cases with occult spinal dysraphism and appear to be the most important clinical predictors (24,28).

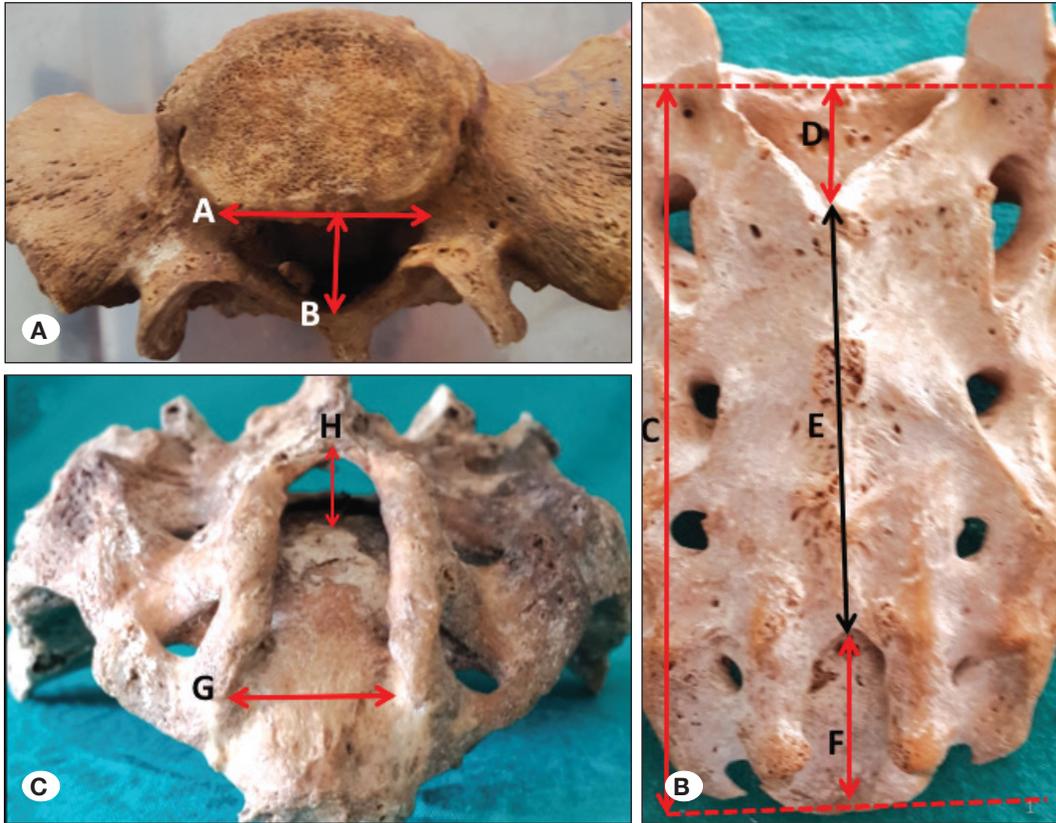
In cases without external manifestations, SBO of the sacrum may cause backache, posterior disc herniation, enuresis, functional disorders of the lower urinary tract, and neurological foot abnormalities (1,18,22). Depending on the level of the lesion, erectile dysfunction is observed in approximately 75% of adult males with spina bifida (14,23). Before performing any surgical procedure on the sacrum (i.e. caudal epidural block, internal fixation via transpedicular and lateral mass screws), the surgeon must consider the congenital defects of the sacrum to help prevent serious surgical complications (61,62,64)

The objective of the present study is to investigate the incidence, types and morphological and morphometric properties of spina bifida on dry sacral bones.

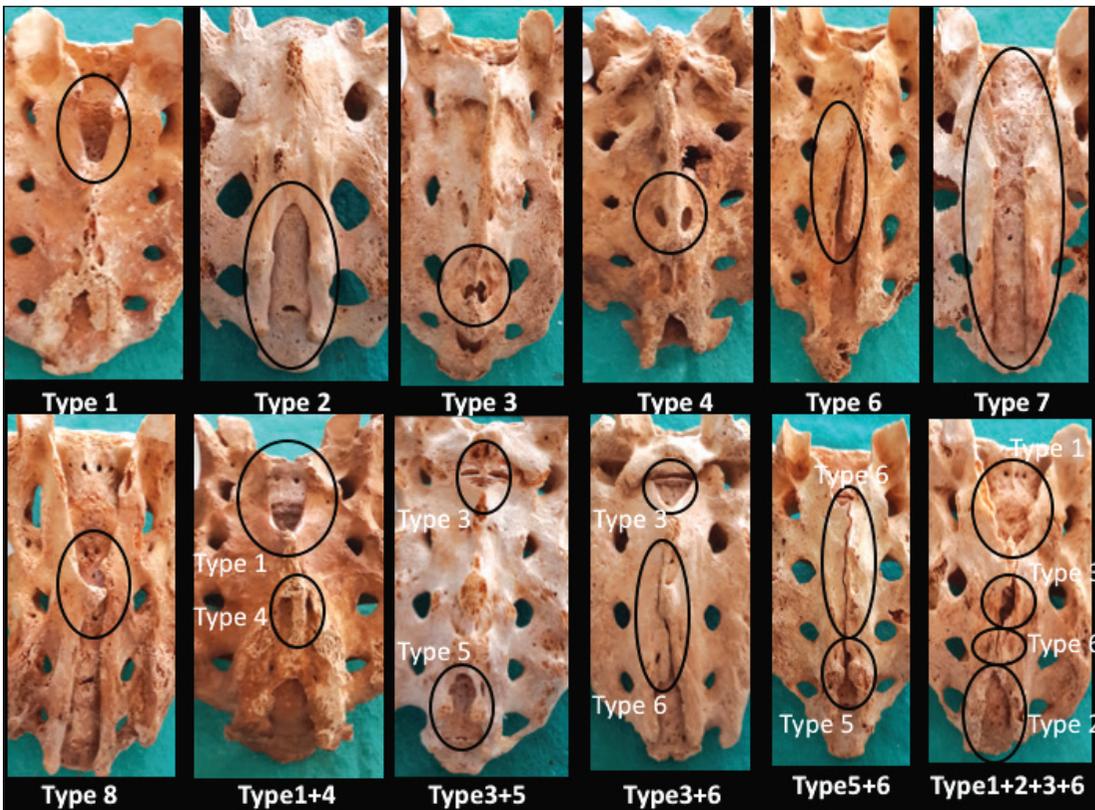
## ■ MATERIAL and METHODS

We examined 110 dry adult sacrum samples gathered from bone samples in the Laboratory of the Anatomy Department of Dokuz Eylul University School of Medicine. The bone samples were obtained from the West Anatolian region. The sex and age of the bones were unknown. Permission for this investigation was obtained from Dokuz Eylul University School of Medicine. The sacrum samples were macroscopically evaluated for the presence of dorsal wall closure defects (spina bifida), as well as the levels and types of these defects.

In the present study, the parameters analysed were: 1) results of parameters related to posterior sacral wall (Table I, Figure 1A-C); 2) classification and rate of closure defects (Table II, Figure 2); 3) classification of the top sacral vertebrae according to the shape of its superior surface (cavity, hump, or flat) (Figure 3A-C); 4) presence and rate of sacralisation and lumbalisation among sacrum samples with dorsal wall defects (Figure 4A, B); 5) vertebral levels of apex of the sacral hiatus (Table III, Figure 5A-F); and 6) vertebral levels of closure defects of the sacrum (Table IV).



**Figure 1:** Measured parameters. **1A)** horizontal (A) and anteroposterior (B) diameter of the sacral canal superiorly, **1B)** posterior height of the sacrum (C), the distance between the median sacral crest and the upper margin of the sacrum (D), the length of the median sacral crest (E), the height of apex of the sacral hiatus (F), **1C)** the distance between sacral corns at sacral hiatus (G), the anteroposterior distance at apex of sacral hiatus (H).



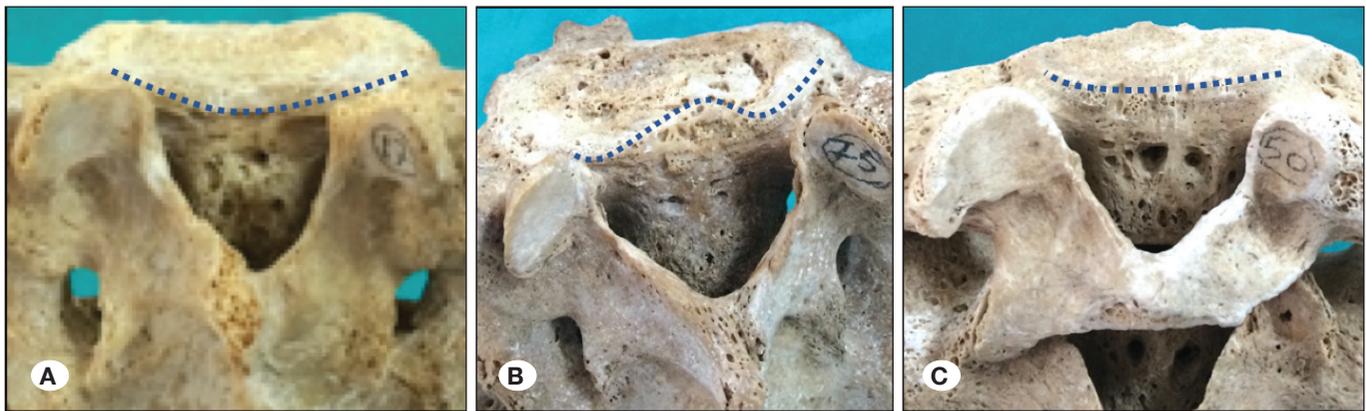
**Figure 2:** The types of posterior sacral wall closed defects. "V" shaped (Type 1), inverse "V" shaped (Type 2), window shaped (Type 3), foramen (hole) shaped (Type 4), sand watch shaped (Type 5), narrow linear (Type 6), wide linear (Type 7), and bridged (Type 8).

**Table III:** The Vertebral Level of Apex of the Sacral Hiatus

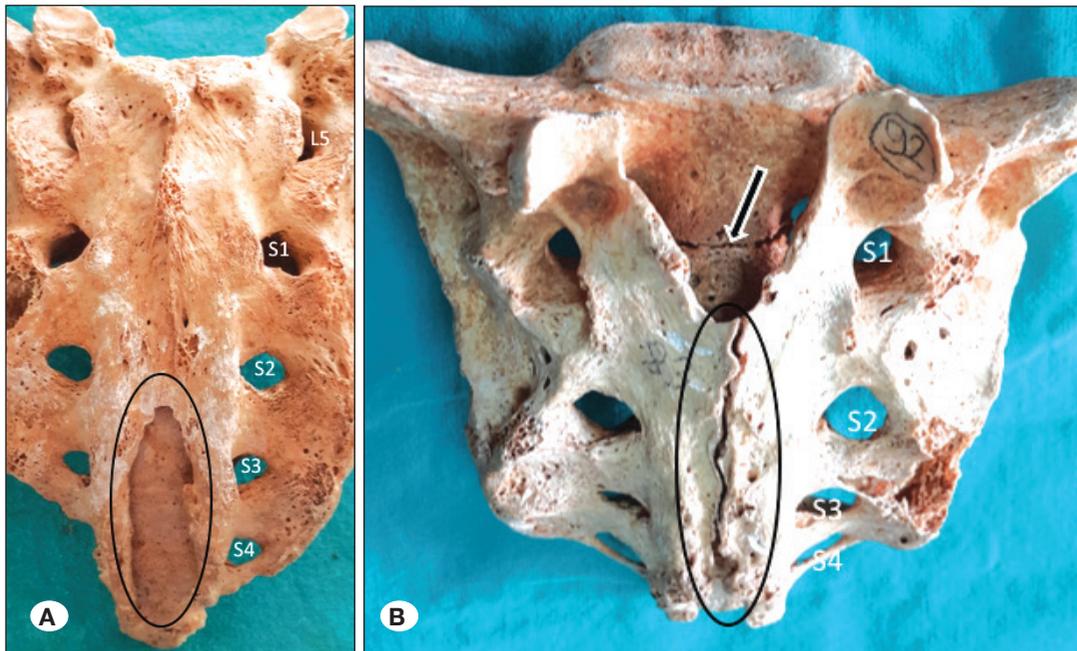
| Level    | Completed Open Posterior Wall | Sacral 2 vertebra | Sacral 3 vertebra | Sacral 4 vertebra | Sacral 5 vertebra | Completed Close Sacral Hiatus |
|----------|-------------------------------|-------------------|-------------------|-------------------|-------------------|-------------------------------|
| Case     | 4                             | 1                 | 7                 | 7                 | 1                 | 2                             |
| N (rate) | (18.18%)                      | (4.54%)           | (31.82%)          | (31.82%)          | (4.54%)           | (9.09%)                       |

**Table IV:** The Vertebral Level of Closure Defect of the Sacrums

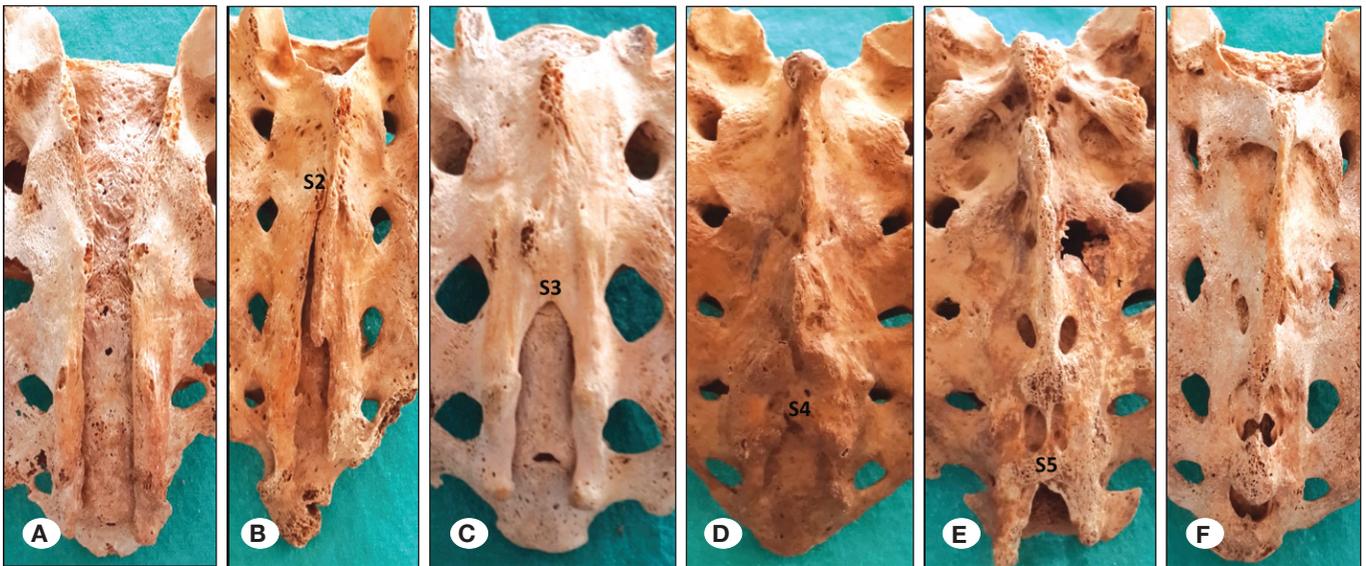
| Level    | L5 vertebra | S1 vertebra | S2 vertebra | S3 vertebra | S4 vertebra | S5 vertebra |
|----------|-------------|-------------|-------------|-------------|-------------|-------------|
| Case     | 3           | 13          | 12          | 14          | 22          | 20          |
| N (rate) | (3.6%)      | (15.5%)     | (14.3%)     | (16.7%)     | (26.2%)     | (23.8%)     |



**Figure 3:** The superior surface of corpus of the first sacral vertebrae were classified according to their shape. **A)** cavity, **B)** hump, **C)** flat.



**Figure 4:** Sacrums representing spina bifida with sacralization or lumbalization. **A)** Sacralization, **B)** Lumbalization.



**Figure 5:** Determined the level of the apex of the sacral hiatus. **A)** Completed Open Posterior Wall, **B)** S2 vertebra level, **C)** S3 vertebra level, **D)** S4 vertebra level, **E)** S5 vertebra level, **F)** Completed Close Sacral Hiatus.

All sacrums were photographed, and measurements were obtained using a Mitotoyo digital calliper sensitive to 0.01 mm.

## RESULTS

We determined that 22 of the 110 sacrums (20%) demonstrated complete (4 of 22 [18.18%]) or incomplete (18 of 22 [81.82%]) spina bifida (Table II). We also observed the coexistence of spina bifida with sacralisation (6 of 22 [27.27%]) and lumbalisation (5 of 22 [22.73%]) (Figure 4A, B). The types of the defects were grouped as 'V' (Type 1), inverse 'V' (Type 2), window (Type 3), foramen (hole) (Type 4), sand watch (Type 5), narrow linear (Type 6), wide linear (Type 7), and bridged (Type 8) (Table II, Figure 2). Types 1, 2, and 3 were the most frequently observed defects in the dorsal wall of the sacrums (4 of 22 [18.18%] for each type). There were 2 cases with a Type 4 (2 of 22 [9.09%]) defect, and 1 case each involving Type 6 (1 of 22 [4.54%]), Type 7 (1 of 22 [4.54%]), and Type 8 (1 of 22 [4.54%]) defects. We observed that Type 5 is not a stand-alone defect, and it was found in 2 cases combined with Type 3 (1 of 22 [4.54%]) and Type 6 (1 of 22 [4.54%]) defects. Additionally, there were 3 other cases with combined defects: Types 1 and 4 (1 of 22 [4.54%]), Types 3 and 6 (1 of 22 [4.54%]), and Types 1, 2, 3, and 6 (1 of 22 [4.54%]) (Table II). There were only 4 sacrums with a completely open posterior sacral wall (Table II). The evaluated parameters were as follows: horizontal (A) and anteroposterior (B) diameter of the sacral canal superiorly, posterior height of the sacrum (C), distance between the median sacral crest and the upper margin of the sacrum (D), length of the median sacral crest (E), height of apex of the sacral hiatus (F), distance between the sacral corns at the sacral hiatus (G), and anteroposterior distance at the apex of the sacral hiatus (H) (Table I, Figure 1A-C). The shape of the upper surfaces of the sacrums (superior surface of the corpus of the S1 vertebrae) with spina bifida

were evaluated and grouped as: cavity (20 of 22 [90.9%]), hump (1 of 22 [4.5%]), or flat (1 of 22 [4.5%]) (Figure 3A-C).

We also determined, in descending order, the location of the apex of the sacral hiatus: S4 level (7 of 22 [31.82%]), S3 level (7 of 22 [31.82%]), S2 level (1 of 22 [4.54%]), and S5 level (1 of 22 [4.54%]). There were 4 cases with a completely open sacral canal, and 2 cases with a completely closed sacral hiatus (Table III, Figure 5A-F).

We observed 6 cases (27.27%) with sacralisation and 5 cases (22.72%) with lumbalisation among the 22 sacrums with closure defects in the dorsal wall (Figure 4A, B).

The location of the different types of dorsal closure defects among the 22 sacrums were determined, in descending order, as follows: S4 level (22 of 22), S5 level (20 of 22), S3 level (14 of 22), S1 level (13 of 22), and S2 level (12 of 22) (Table IV).

## DISCUSSION

Spina bifida, is a group of developmental anomalies which occur as a result of defects in neural tube closure (76). With an occurrence rate of 1 per every 1000 pregnancies preceding folic acid fortification, neural tube defects are the second most prevalent malformation after congenital heart defects (11). De Bakker et al. declared that the human neural tube unites at a single site and that neural tube defects are not limited to a specific location (13).

Spinal dysraphism (Greek: bad suture) defines all types of congenital spinal disorders that involve abnormal differentiation or incomplete closure defect at the midline of the mesenchymal, osseous, and neural tissue (45,68). The severity of the condition is directly influenced by whether the nervous tissue or meninges are protected by skin or whether they are exposed to the environment (36).

The spina bifida cystica refers to a neural tube defect, whereas SBO refers to a small defect brought about by the nonclosure of the posterior vertebral neural arches (36). Paleopathologists have mostly accepted and reported that SSBO was a congenital anomaly (e.g. 15,17,53,71). However, there is no consensus on the level of closure defects that should be considered as SSBO. Under normal conditions, the neural arches of the last 2 sacral segments remain underdeveloped, thus forming the sacral hiatus (6,12).

There are variations at the level of nonclosure of the lamina of the sacral bodies (56). Normal variations of the sacral hiatus are observed at the S4–S5 vertebrae levels with high frequency and at the S3 level with intermediate frequency. Therefore, SSBO that is determined at the S3, S4, or S5 level is accepted as being within the normal variation (e.g.1,19,26,43,53). Bagheri and Govsa examined 87 dry adult human sacrum and determined the rate spina bifida rate to be 19% at the S1–S5 level (5). In addition to the above-mentioned studies, we determined this rate as 20% (22 of 110). In the present study, the SSBO were observed at the S1 level in 13 sacrum (11.82%) (Table IV).

We also grouped the sacrum with SSBO according to the shape of the closure defects: 'V', inverse 'V', window, foramen (hole), sand watch, narrow linear, wide linear, bridged (Table II, Figure 2). The most frequently observed shapes were 'V' (4 of 22 [18.18%]), inverse 'V' (4 of 22 [18.18%]), and window (4 of 22 [18.18%]). Singh and Mahajan examined 159 dry human sacrum and also grouped the shapes of the sacral hiatus using the following terms: inverted 'U' (42.95%), inverted 'V' (27.51%), irregular (16.10%), dumbbell (11.40%), and bifid (2.01%) (63). Similar to Singh and Mahajan (63), Bagheri and Govsa (5) examined 87 adult dry sacrum according to the shape of the sacral hiatus and grouped them as: inverted 'U' (33.33%), inverted 'V' (19.45%), irregular (19.45%), dumbbell (6.90%), bifid (3.45%), and 'M' shaped (10.34%). In these studies, the types of closure defects were described and grouped differently and, therefore, the rates of the each type of closure defect have been observed differently.

Tardieu et al. reported that Muscatello investigated patients with SBO and found that it was most commonly observed in the caudal part of the spine (66). French reported the incidence of SBO in adult cases as approximately 10% and, similar to Muscatello's findings, commonly encountered in the lumbosacral region (21). In the present study, the incidence of spina bifida (complete and incomplete) was observed as 20% (22 of 110). SSBO in adults may not cause any symptoms for many years and may, therefore, be diagnosed coincidentally. This may be the reason why the incidence is twice as common in the present study than what has been reported in the normal population.

Eubanks and Cheruvu studied 3100 dry sacrum and reported that the defect extended from S1 to S5 in only 12.4% of SSBO cases, meaning that total SSBO was found in only 1.2% of the population studied (16). Kumar et al. studied dry bone sacrum in India and observed the deficient dorsal wall of the sacrum as 2.7% and 12.5% in males and females, respectively (38). In the present study, we determined that 22 of the 110 sacrum

(20%) had spina bifida, 4 (18.18%) with complete and 18 (81.82%) with incomplete spina bifida (Table II). Eubanks and Cheruvu examined a large collection of sacrum from both white and black races and found the incidence of spina bifida to be 1.2% (16). This difference may be the result of geographic conditions, racial differences or number of bones evaluated in the studies.

Seçer et al. evaluated 401 patients with low back pain (LBP) lasting longer than 2 weeks by using standard lumbosacral x-rays. They found that 34 (8.5%) patients had spina bifida, which was most commonly observed at the level of the S1 vertebra (30 patients [7.48%]) (60). In this study, 52 (12.1%) of the 401 patients with LBP were reported to have congenital vertebral abnormalities. Of the congenital vertebral abnormalities, 34 (8.5%) were spina bifida and 18 (4.5%) were transitional vertebra (60). In the current study, we determined the number of sacrum with spina bifida as 22 of the 110 (20%) examined. Of these, 11 (50%) had transitional vertebra. Six cases (27.27%) had sacralisation and 5 cases (22.72%) had lumbalisation defects (Figure 4A, B). The rate of spina bifida was higher in our study than the rates reported previously. This difference may be due to the differences in the studied material and population. Seçer et al. performed their study using lumbosacral x-rays of patients with LBP and we evaluated the human dry sacrum (60).

We measured the posterior length of the sacrum as  $113.0 \pm 11.21$  mm. Maddikunta and Ravinder studied 60 adult sacrum (27 male, 33 female) and measured the posterior length of sacrum as  $113.9 \pm 11.53$  mm in males and  $90.0 \pm 9.13$  mm in females (41). Ravichandran et al. studied 123 sacrum (63 males, 60 females) and measured the mean posterior length as 97.8 mm in males and 90.96 mm in females (54). Both of these studies were performed in India, but in different geographic regions. The length of the sacrum in males was longer in Ravichandran et al.'s (54) study compared with that of Maddikunta and Ravinder (41). This difference may be due to the differences in sample size. Furthermore, regional differences may account for the differences in length. The results of male sacrum examinations in Maddikunta and Ravinder's (41) study were almost the same as the results of the present study, but we do not have any knowledge regarding the sex of the bones.

Developmental errors of vertebrae are highly variable, ranging from 1 level to multiple defects throughout the spine, and may affect any part of the vertebrae (neural arch defects, segmental defects of vertebral bodies, anterior vertebral body defects, and minor fusion anomalies) (74). The vertebral defects that cause congenital scoliosis may be caused by failure of formation, failure of segmentation or a combination of both (3,25,27,29,72). Cowell and Cowell studied the coexistence of SBO and idiopathic scoliosis in 100 patients and observed the incidence as 34% (3 at L5, 31 at S1) (12). In a recent study, Passias et al. reported clinical data for the coexistence of spinal anomalies, such as spina bifida and scoliosis. They reported that 11.4% of patients with spinal anomalies had scoliosis (50). Marchetti and Bartolozzi categorised spondylolisthesis as developmental

spondylolisthesis and acquired spondylolisthesis (42). According to the severity of the bony dysplastic changes, developmental spondylolisthesis was further divided into 2 groups: low dysplastic and high dysplastic. High dysplastic spondylolisthesis has a congenital nature and is found in one-third of the patients with SBO of either L5, the sacrum or both (4,75). High grade L5-S1 spondylolisthesis is associated with a variable degree of bony dysplasia, including rounding of the upper sacral endplate (sacral doming), an abnormal superior sacral facet, malformations such as hypoplasia of the upper surface of the body of the first sacral vertebra, hypoplasia or aplasia of the facets, and other dysplastic changes (70). The coexistence of these malformations with spina bifida reduces the efficiency of the stabilising system represented by the posterior articular complex (40). Therefore, the definition of the upper sacral endplate is of importance. In the present study, we evaluated the upper sacral surfaces of sacrums with spina bifida. We did not observe any dome-shaped upper surfaces. Furthermore, we noted the shape of the upper surfaces as 20 (90.9%) cavity, 1 (4.5%) hump, and 1 (4.5%) flat (Figure 3A-C).

Bony defects of the sacrum may be accompanied by fibrous bands, fatty or other tumours occupying the area on the laminal defect. This can cause compression of the nerves of the cauda equina, resulting in interference in nerve conduction in the sacral reflex arcs (32,39,65).

Depending on the level and nature of the lesions that affect the spinal cord in spina bifida patients with myelomeningocele, serious motor, sensory and autonomic disorders can occur in the lower limbs, bladder, and bowels (7). Therefore, faecal incontinence, constipation, and urinary incontinence and retention are major problems profoundly affecting the life quality and social integration of spina bifida patients (67). Galloway and Tainsh reported an increased prevalence of SBO among adults with lower urinary tract problems (22). The incidence of SBO was determined to be 35% to 60% among enuretic children (34,37,55,57). However, the rate was also as high as 17% in normal children (8). Kumar et al. reported the outcome of enuresis treatment in children with SBO to be relatively the same as that for normal children (37). Pippi Salle et al. found SBO in 70% of children with voiding dysfunction (52).

The abnormality presents clinical importance with respect to caudal epidural block (CEB), which is used in the diagnosis and treatment of lumbar spine disorders (61). The anatomical variations may lead to the failure of the CEB or transpedicular and lateral mass screw placement (61,64). Spina bifida may lead to a possible increased risk of damage to the sacral nerves and present difficulty in internal fixation by screws (64). Knowledge about the anatomic distance between the sacral hiatus and dural sac is important in CEB with respect to the risk of dural puncture. The causes of failure of CEB are bony septum in the sacral hiatus, hiatal agenesis, or complete agenesis (spina bifida) in 7% of cases. Additionally, in 1% of cases, total spina bifida and detection of the dura mater just beneath the hiatus were reported (61,62). Park et al. applied caudal epidural steroid injection to patients in Korea using ultrasound guidance and measured the intercornual distance

between the apex of both cornua as  $16.9 \pm 2.7$  mm (49). In the present study, we measured the mean distance between the bilateral cornua and found similar results ( $16.47 \pm 2.90$  mm). Bagheri and Govsa examined 87 dry adult human sacral bones and measured the length of sacral hiatus as  $28.07 \pm 7.1$  mm (5). Singh and Mahajan measured the mean length of sacral hiatus as 22.69 mm in India, whereas we measured the length of sacral hiatus as  $24.51 \pm 11.30$  mm (63). This difference may be because of the cases with spina bifida included in our study. We determined mostly the location of the apex of the sacral hiatus at S4 and S3 level (14 of 22 [63,64%]). Similarly, Kumar et al. found the location of the apex at S4 in 76.23% of cases (38). At the level of apex, Pal et al. measured the mean anteroposterior diameter of sacral canal as 5.34 mm, whereas we measured it as  $5.63 \pm 2.51$  mm (47).

## ■ CONCLUSION

It is essential to precisely define the anatomical variations of sacrums for surgeons, particularly when they are operating using endoscopic techniques, and for anaesthesiologists applying CEB. We assume that the present study may guide medical doctors and surgeons during diagnostic and surgical procedures related to the sacrum.

## ■ REFERENCES

1. Albrecht TL, Scutter SD, Henneberg M: Radiographic method to assess the prevalence of sacral spina bifida occulta. *Clin Anat* 20:170-174, 2007
2. Aquilera S, Soothill P, Denbow M, Pople I: Prognosis of spina bifida in the era of prenatal diagnosis and termination of pregnancy. *Fetal Diagn* 26:68, 2009
3. Arlet V, Odent T, Aebi M: Congenital scoliosis. *Eur Spine J* 12:456-463, 2003
4. Babbi L, Terzi S, Bandiera S, Barbanti Brodano G: Spina bifida occulta in high grade spondylolisthesis. *European Review for Medical and Pharmacological Sciences* 18 Suppl 1:8-14, 2014
5. Bagheri H, Govsa F: Anatomy of the sacral hiatus and its clinical relevance in caudal epidural block. *Surg Radiol Anat* 39:943-951, 2017
6. Barnes E: Developmental defects of the axial skeleton in paleopathology. Colorado: University Press of Colorado, 1994: 360
7. Behrman RE, Kliegman RM, Jenson HB: Disorders of the central nervous system. In: Nelson Textbook of Paediatrics. 16<sup>th</sup> ed. Philadelphia: WB Saunders, 2000
8. Boone D, Parsons D, Lachmann SM, Sherwood T: Spina bifida occulta: Lesion or anomaly? *Clin Radiol* 36:159-161, 1985
9. Brinker MR, Rosenfeld SR, Feiwell E, Granger SP, Mitchell DC, Rice JC: Myelomeningocele at the sacral level. Long-term outcomes in adults. *J Bone Joint Surg Am* 76(9):1293-1300, 1994
10. Cameron M, Moran P: Prenatal screening and diagnosis of neural tube defects. *Prenat Diagn* 29:402-411, 2009
11. Copp AJ, Greene ND, Murdoch JN: The genetic basis of mammalian neurulation. *Nat Rev Genet* 4:784-793, 2003

12. Cowell MJ, Cowell HR: The incidence of spina bifida occulta in idiopathic scoliosis. *Clin Orthop Relat Res* 118:16-18, 1976
13. De Bakker BS, Driessen S, Boukens BJD, van den Hoff MJB, Oostra RJ: Single-site neural tube closure in human embryos revisited. *Clin Anat* 30:988-999, 2017
14. Diamond DA, Rickwood AM, Thomas DG: Penile erections in myelomeningocele patients. *Br J Urol* 58:434-435, 1986
15. El-Din AMS, El-Banna RA: Congenital anomalies of the vertebral column: A case study on ancient and modern Egypt. *Int J Osteoarchaeol* 16:200-207, 2004
16. Eubanks JD, Cheruvu VK: 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. *Spine (Phila Pa 1976)* 34(15):1539-1543, 2009
17. Ferembach D: Frequency of spina bifida occulta in prehistoric human skeletons. *Nature* 199:100-101, 1963
18. Fidas A, MacDonald HL, Elton RA, McInnes A, Wild SR, Chisholm GD: Prevalence of spina bifida occulta in patients with functional disorders of the lower urinary tract and its relation to urodynamic and neurophysiological measurements. *BMJ* 298:357-359, 1989
19. Fidas A, MacDonald HL, Elton RA, Wild SR, Chisholm GD, Scott R: Prevalence and patterns of spina bifida occulta in 2707 normal adults. *Clin Radiol* 38:537-542, 1987
20. Findley TW, Agre JC, Habeck RV, Schmalz R, Birkebak RR, McNally MC: Ambulation in the adolescent with myelomeningocele. I: Early childhood predictors. *Arch Phys Med Rehabil* 68:518-522, 1987
21. French BN: Midline fusion defects and defects of formation. In: Youmans JR (ed), *Youmans Neurological Surgery*, 3<sup>rd</sup> ed. Philadelphia: WB Saunders, 1990:1081-1235
22. Galloway NT, Tainsh J: Minor defects of the sacrum and neurogenic bladder dysfunction. *Br J Urol* 57:154-155, 1985
23. Gamé X, Moscovici J, Gamé L, Sarramon JP, Rischmann P, Malavaud B: Evaluation of sexual function in young men with spina bifida and myelomeningocele using the International Index of Erectile Function. *Urology* 67:566-570, 2006
24. Guggisberg D, Hadj-Rabia S, Viney C, Bodemer C, Brunelle F, Zerah M, Pierre-Kahn A, de Prost Y, Hamel-Teillac D: Skin markers of occult spinal dysraphism in children. *Arch Dermatol* 140:1109-1115, 2004
25. Hedequist D, Emans J: Congenital scoliosis. A review and update. *J Pediatr Orthop* 27(1):106-116, 2007
26. Henneberg RJ, Henneberg M: Variation in the closure of the sacral canal in the skeletal sample from Pompeii, Italy, 79 AD. *Perspect Hum Biol* 4:177-188, 1999
27. Hensinger RN: Congenital scoliosis aetiology and associations. *Spine* 34(17):1745-1750, 2009
28. Henriques JG, Pianetti G, Henriques KS, Costa P, Gusmao S: Minor skin lesions as markers of occult spinal dysraphisms e prospective study. *Surg Neurol* 63 Suppl 1:8-12, 2005
29. Herman JM, McLone DG, Storrs BB, Dauser RC: Analysis of 153 patients with myelomeningocele or spinal lipoma reoperated upon for a tethered cord. *Pediatr Neurosurg* 9:243-249, 1993
30. Hoffer MM, Feiwell E, Perry R, Perry J, Bonnett C: Functional ambulation in patients with myelomeningocele. *J Bone Joint Surg Am* 55(1):137-148, 1973
31. Holmbeck GN, Faier-Routman J: Spinal lesion level, shunt status, family relationships, and psychosocial adjustment in children and adolescents with spina bifida myelomeningocele. *Journal of Pediatric Psychology* 20:817-832, 1995
32. James CCM, Lassman LP: *Spina bifida occulta*. London: Academic Press 1981: 215-219
33. Kamanli A, Genc H: Radiological abnormalities of the lumbosacral spine in young male individuals. *J Back Musculoskelet Rehabil* 16(2):91-94, 2002
34. Kawauchi A, Kitamori T, Imada N, Tanaka Y, Watanabe H: Urological abnormalities in 1,328 patients with nocturnal enuresis. *Eur Urol* 29:231-234, 1996
35. Kondo A, Kamihira O, Ozawa H: Neural tube defects: Prevalence, etiology and prevention. *Int J Urol* 16:49-57, 2009
36. Kumar A, Tubbs RS: Spina bifida: A diagnostic dilemma in paleopathology. *Clin Anat* 24(1):19-33, 2011
37. Kumar P, Aneja S, Kumar R, Taluja V: Spina bifida occulta in functional enuresis. *Indian J Pediatr* 72:223-225, 2005
38. Kumar V, Nayak SR, Potu BK, Pulakunta T: Sacral hiatus in relation to low back ache in south Indian population. *Bratisk Lek Listv* 110(7):436-441, 2009
39. Lemire RJ, Beckwith JB: Pathogenesis of congenital tumors and malformations in the sacrococcygeal region. *Teratology* 25:201-213, 1982
40. Logroscino G, Mazza O, Aulisa G, Pitta L, Pola E, Aulisa L: Spondylolysis and spondylolisthesis in the pediatric and adolescent population. *Childs Nerv Syst* 17(11):644-655, 2001
41. Maddikunta V, Ravinder M: Role of morphometrical data of sacrum in determination of sex. *Int J Biol Med Res* 5(1):3785-3792, 2014
42. Marchetti PG, Bartolozzi P: Classification of Spondylolisthesis as a Guideline for Treatment. *The Textbook of Spinal Surgery*, 2<sup>nd</sup> ed. Lippincott-Raven Publishers, Philadelphia, 1997:1211-1254
43. Masnicova S, Benus R: Developmental anomalies in skeletal remains from the Great Moravia and Middle Ages cemeteries at Devin (Slovakia). *Int J Osteoarchaeol* 13:266-274, 2003
44. McDonald CM, Jaffe KM, Mosca VS, Shurtleff DB: Ambulatory outcome of children with myelomeningocele: Effect of lower-extremity muscle strength. *Dev Med Child Neurol* 33(6):482-490, 1991
45. Naidich TP, McLone DG, Harwood-Nash DC: Spinal dysraphism. In: Newton TH, Potts DG (eds), *Modern Neuroradiology*, Vol 1: Computed Tomography of the Spine and Spinal Cord. San Anselmo: Clavadel Press, 1983:299-353
46. Ong LC, Lim YN, Sofiah A: Malaysian children with spina bifida: Relationship between functional outcome and level of lesion. *Singapore Medical Journal* 43:12-17, 2002
47. Pal DR, Rahman MA, Fatema K: Morphometric study of sacral hiatus: A basis for successful caudal epidural block. *Bangladesh Journal of Anatomy* 10(1):5-10, 2012

48. Papp T, Porter RW: Changes of the lumbar spinal canal proximal to spina bifida occulta. An archaeological study with clinical significance. *Spine* 19:1508-1511, 1994
49. Park GY, Kwon DR, Cho HK: Anatomic differences in the sacral hiatus during caudal epidural injection using ultrasound guidance. *Journal of Ultrasound in Medicine* 34(12):2143-2148, 2015
50. Passias PG, Poorman GW, Jalai CM, Diebo BG, Vira S, Horn SR, Baker JF, Shenoy K, Hasan S, Buza J, Bronson W, Paul JC, Kaye I, Foster NA, Cassilly RT, Oren JH, Moskovich R, Line B, Oh C, Bess S, LaFage V, Errico TJ: Incidence of congenital spinal abnormalities among pediatric patients and their association with scoliosis and systemic anomalies *J Pediatr Orthop* 39(8):608-613, 2019
51. Pierre-Kahn A, Zerah M, Renier D, Cinalli G, Sainte-Rose C, Lellouch-Tubiana A, Brunelle F, Le Merrer M, Giudicelli Y, Pichon J, Kleinknecht B, Nataf F: Congenital lumbosacral lipomas. *Childs Nerv Syst* 13(6):298-334, 1997
52. Pippi Salle JL, Capolicchio G, Houle AM, Vernet O, Jednak R, O'Gorman AM, Montes JL, Farmer JP: Magnetic resonance imaging in children with voiding dysfunction: Is it indicated? *J Urol* 160:1080-1083, 1998
53. Post RH: Pilot study: Population differences in the frequency of spina bifida occulta. *Eugen Q* 13:341-352, 1966
54. Ravichandran D, Shanthy KC, Shankar, K, Chandra H: A study on sacral index in Tamil Nadu and Andhra Pradesh population of Southern India. *J Clin Diagn Res* 7(9):1833-1834, 2013
55. Ritchey ML, Sinha A, DiPietro MA, Huang C, Flood H, Bloom DA: Significance of spina bifida occulta in children with diurnal enuresis. *J Urol* 152:815-818, 1994
56. Romanes GJ: *Cunningham's Textbook of Anatomy*. 12th ed., Oxford: Oxford University Press, 1981:626-639
57. Samuel M, Boddy SA: Is spina bifida occulta associated with lower urinary tract dysfunction in children? *J Urol* 171:2664-2666, 2004
58. Schoenmakers M, Gulmans V, Gooskens R, Helden P: Spina bifida at the sacral level: More than minor gait disturbances. *Clinical Rehabilitation* 18:178-185, 2004
59. Schwartz S, Cohen ME, Herbison GJ, Shah A: Relationship between two measures of upper extremity strength: Manual muscle test compared to hand-held myometry. *Arch Phys Med Rehabil* 73(11):1063-1068, 2004
60. Secer M, Muradov JM, Dalgic A: Evaluation of congenital lumbosacral malformations and neurological findings in patients with low back pain. *Turk Neurosurg* 19(2):145-148, 2009
61. Sekiguchi M, Yabuki S, Satoh K, Kikuchi S: An anatomic study of the sacral hiatus: A basis for successful caudal epidural block. *Clin J Pain* 20:51-54, 2004
62. Senoglu N, Senoglu M, Gumusalan Y: Total spina bifida occulta of the sacrum. *IJAV* 1:27-28, 2008
63. Singh M, Mahajan A: An anatomical study of variations of sacral hiatus in sacra of North Indian origin and its clinical significance. *Int J Morphol* 31(1):110-114, 2013
64. Srijit D, Shipra P: Spina bifida with higher position of sacral hiatus: a case report with clinical implications. *Bratisl Lek Listy* 108:467-469, 2007
65. Sutow WW, Pryde AW, Kastenbaum MA: Incidence of spina bifida occulta in relation to age. *AMA J Dis Child* 91(3):211-217, 1956
66. Tardieu GG, Loukas M, Fisahn C, Shoja MM, Oskouian RJ, Tubbs RS: The Italian Giuseppe Muscatello (1866-1951) and his contributions to our understanding of childhood spina bifida aperta and occulta. *Childs Nerv Syst* 33(3):389-391, 2017
67. Tjandra JJ, Ooi BS, Han WR: Anorectal physiologic testing for bowel dysfunction in patients with spinal cord lesions. *Dis Colon Rectum* 43:927-931, 2000
68. Tortori-Donati P, Rossi A, Cama A: Spinal dysraphism: A review of neuroradiological features with embryological correlations and proposal for a new classification. *Neuroradiology* 42:471-491, 2000
69. Turkel SJ: Congenital abnormalities in skeletal populations. In: Iscan MY, Kennedy KAR, (eds), *Reconstruction of Life from the Skeleton*. New York: AR Liss, 1989:109-127
70. Wang Z, Parent S, de Guise JA, Labelle H: A variability study of computerized sagittal sacral radiologic measures. *Spine* 35(1):71-75, 2009
71. Webb S: *Paleopathology of Aboriginal Australians: Health and Disease Across a Hunter-Gatherer Continent*. Cambridge: Cambridge University Press, 1995: 336
72. Winter RB: Congenital spinal deformity. Lonstein JE, Bradford DS, Winter RB, Ogilvie JW (eds), *Moe's Textbook of Scoliosis and Other Spinal Deformities*. Philadelphia:Saunders, 1995:3
73. Wolf LS, McLaughlin JF: Early motor development in infants with meningomyelocele. *Pediatric Physical Therapy* 4:12-17, 1992
74. Wynne-Davies R: Congenital vertebral anomalies: Aetiology and relationship to spina bifida cystica. *J Med Genet* 12(3):280-288, 1975
75. Wynne-Davies R, Scott JH: Inheritance and spondylolisthesis: A radiographic family survey. *J Bone Joint Surg Br* 61-B(3):301-305, 1979
76. Yen IH, Houry MJ, Erickson JD, James LM, Waters GD, Berry RJ: The changing epidemiology of neural tube defects. United States, 1968-1989. *Am J Dis Child* 146: 857-861, 1992