



# Complete Dorsal Wall Agenesis of the Sacral Canal in a Greek Population: an Osteological Study

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## Abstract

**Introduction:** The failure of closure of the dorsal wall of the sacral canal (SC) has been known since the eve of modern osteology, appearing in prehistoric times. Variants include partial or complete absence of the dorsal wall of the SC. SC presents a pathway for minimally invasive therapeutic and diagnostic procedures for spinal diseases and for ensuring analgesia and anesthesia in operations, including labor and genitourinary surgery.

**Aim:** Our objective is to verify the incidence of complete agenesis of the SC dorsal wall in Greek population.

**Material and methods:** We collected 155 adult dry sacra of known sex from the Third Cemetery of Athens for the study of the dorsal wall of the sacrum. Damaged or variated sacra were excluded. We also performed an exhaustive review of the relevant literature and compared our results with those of international studies.

**Results:** Complete dorsal wall agenesis of the sacral canal was found in three cases (1.93%), two men (1.29%) and one woman (0.64%). Our review examines its incidence in other populations and explores the possibility of regional or racial correlation.

**Conclusion:** Knowing the complete dorsal wall agenesis of the sacral canal by medical professionals is crucial for avoiding complications in spinal surgery, anesthetics and obstetrics, as well as in the differential diagnosis of neurological and urological diseases.

## Keywords

complete agenesis, Greek, sacrum, sacral variations, sacral canal, spina bifida

## INTRODUCTION

The sacral canal (SC) is the peripheral part of the vertebral canal, extending from the level of S1 vertebra to the sacral hiatus. Since the spinal cord terminates at approximately L2, SC encloses the cauda equina, including the filum terminale and the spinal meninges. The dura and arachnoid mater typically terminate at the level of S2, but variations include the lower border of S1 foramen in adults and the S3

in children. At the end point, they fuse into one layer, while the pia mater progresses to the coccyx as filum terminale. The sacral canal also contains epidural fat, which becomes denser with age, as well as the valveless sacral epidural vein plexus. This usually ends at S4, though it may extend throughout the canal.<sup>[1]</sup>

Numerous divergences occur in the SC dorsal wall including complete agenesis (total sacral spina bifida).<sup>[2]</sup> Spina bifida is a developmental defect of the neural tube, resulting from

inadequate closure during the early embryonic period. Spina bifida may be divided into two types, a) spina bifida occulta (SBO) in which the meninges and/or neural tissue are hidden, covered by intact skin and b) spina bifida cystica (SBC), which involves the vertebral arches and the meninges, exposing the neural tissue. SBO occurs most commonly in the sacral region. It varies from partial defect of the posterior arch of some vertebrae to pan-sacral S1-S5 defect and has been termed sacral spina bifida occulta (SSBO).<sup>[3]</sup> Singh in 2013 classified SSBO according to the degree of the sacral canal closure impairment in four categories. According to this classification, complete aggenesis of the dorsal wall of the sacral canal is classified as type 1 SSBO.<sup>[4]</sup>

Most cases of spina bifida are of multifactorial origin, influenced by both genetic and environmental factors. Increased risk of spina bifida presents with high pregnancy weight, antiepileptic drugs (valproic acid), folic acid antagonists, maternal diabetes, maternal smoking, hyperthermia, and fever during pregnancy.<sup>[5]</sup> Genetic factors remain elusive even though Singh noted that factors responsible for this condition are the mutant expression of Hox-11, Pax-1/Pax-9, SHH, BMP, Wnt, and Fox2 genes.<sup>[4]</sup> Recent research provides convincing support that the main Hox genes responsible for determining sacral vertebrae are Hox-10 and Hox-11.<sup>[6]</sup> However, Hox genes are not the only contributing factor for normal sacral development. The resegmentation process is regulated by the paired box genes, Pax-1 and Pax-9.<sup>[7]</sup> Finally, other genes such as the planar cell polarity genes (PCP), VANGL1 and CELSR1 have been studied in spina bifida cohorts among Italians, Americans, and the French.<sup>[8]</sup>

## AIM

We conducted a descriptive osteological study aiming to estimate the incidence of complete dorsal wall aggenesis in Hellenic population.

## MATERIALS AND METHODS

We studied 155 dry adult sacra of Hellenic (Caucasian) origin and known sex, retrieved by permission from the authorities of the Third Cemetery of Athens, Greece. Only intact bones were included in the study; sacra with any sign of fracture or variation, e.g. features of sacralization or lumbarization, were excluded. We took photographs of the bones using a digital camera (Nikon DSLR D5300) and saved them in JPEG format. We additionally performed a review of the relevant literature, to compare our findings with the recorded incidence of dorsal wall aggenesis of the sacral canal in diverse populations.

## RESULTS

Complete aggenesis of the dorsal wall was identified in 3 cases of the 155 sacra (1.93%). Two bones belonged to male (1.29%) (Figs 1, 2) and one to female (0.64%) skeletons (Fig. 3). In the female sacrum, coccyx sacralization was evident, with unilateral fusion of the transverse process of the first coccygeal vertebra to the inferolateral angle of the sacrum. SC frontal wall presented no features of variability or abnormality.



**Figure 1.** Case 1 (male).



**Figure 2.** Case 2 (male).



**Figure 3.** Case 3: complete dorsal wall agenesis of sacral canal with unilateral coccyx sacralization (female).

## HISTORICAL PEARLS

SBO appeared in humans during the prehistoric period, evident in skeletal remains.<sup>[9]</sup> Anatomy textbooks of the early

19th century mention the absence of the SC dorsal wall, testifying that this sacrum variation was not only known but considered normal and benign.<sup>[10]</sup> Ernst Ziegler named the median cleft of the SC dorsal wall *rhachischisis* and considered it a

malformation.<sup>[11]</sup> Rudolph Virchow (1821-1902) was the first to describe SBO in a few cases.<sup>[12,13]</sup> Camille Dareste in 1877 presented such cases in his work “La production artificielle Des Mostruosités”.<sup>[14]</sup> Heinrich Hermann Robert Koch (1843-1910) suggested that shortening of the spinal column, the absence of some vertebrae and dwarfing should be considered factors leading to SBO.<sup>[12]</sup> SBO and SBC were in vogue at the end of the second half of the 19th century, connected sometimes with other monstrous deformities. Both SBC and SBO have been described and illustrated in James Morton’s work “The Treatment of spina bifida by a new method”.<sup>[15]</sup> SBO was considered in the late 19th and early 20th centuries as a developmental defect in the closure of the vertebral lamina with or without protrusion of the membranes.<sup>[16]</sup>

## DISCUSSION

### General notes

Dorsal aggenesis of the sacrum (DAS) has been studied in different countries such as the United States of America,

Turkey, Japan, Nigeria, Thailand, India, and others. This is the first time the occurrence of complete aggenesis of the SC dorsal wall is investigated in the Hellenic population. Our aim was to find the incidence of DAS, compare it with the results from other regions and find possible correlations or deviations (**Table 1**).

The incidence of DAS in the literature ranges from 0.43% (Thailand) to 5% (Bangladesh). In our review, the mean incidence out of 35 studies from 1944 to 2023 was approximately 1.97% (**Table 1**). Most of the studies were performed in India (22/35), maybe because of cultural reasons, as the term sacrum continues to have a near mystical status in many cultures.

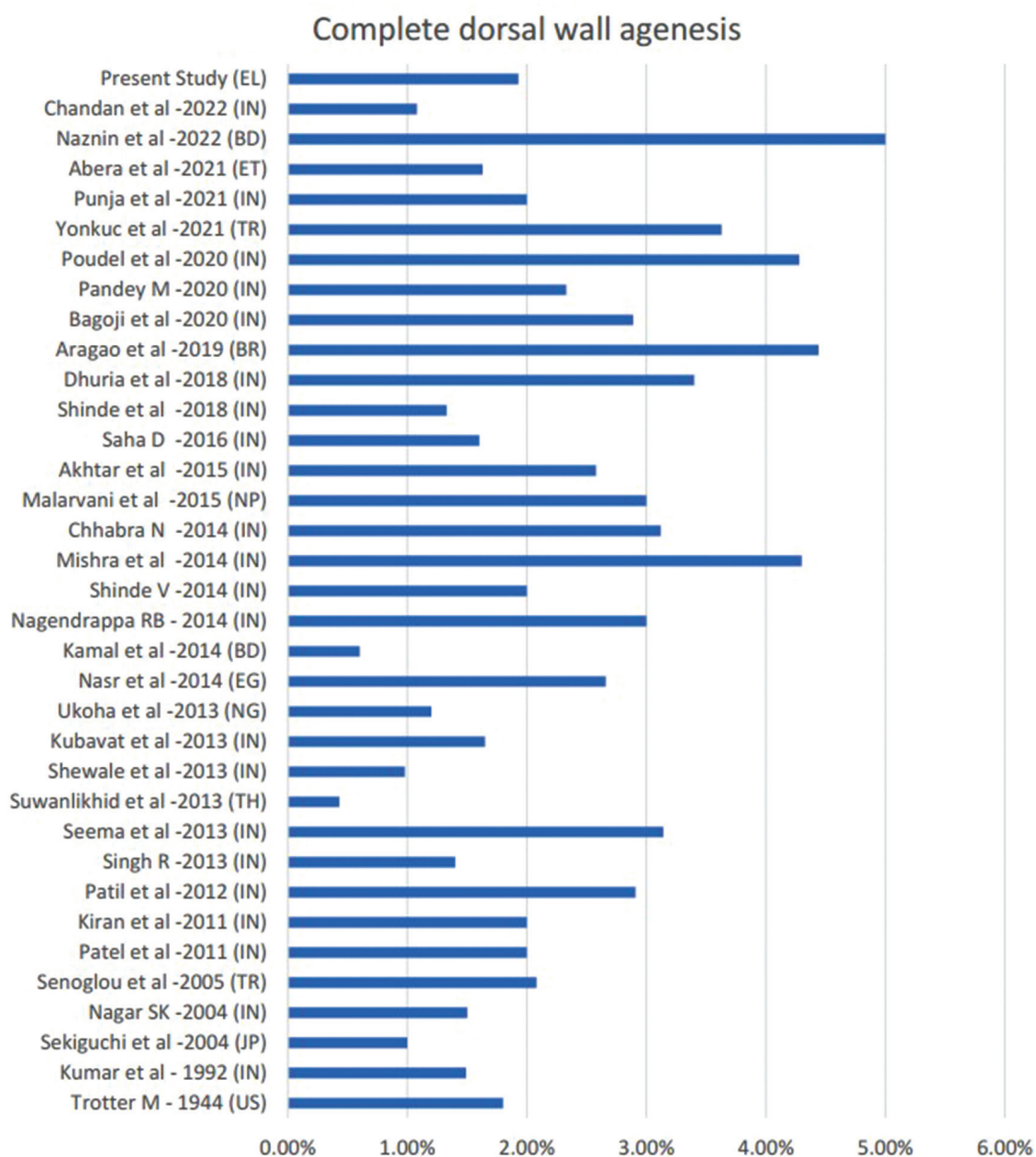
The incidence of DAS in our study was 1.93%, just below average and closest to Senoglou (Turkey) and Patel, Kiran and Shinde (India) findings (**Fig. 4**). In a recent study, Wu et al. reported that the rate of Chinese people with SBO was significantly higher than that among the English, indicating that SBO might be region- or race-dependent. Moreover, they reported the incidence of SBO being significantly higher among men than women.<sup>[50]</sup>

**Table 1.** Incidence of complete dorsal wall aggenesis of the sacral canal in different population groups

No	First author	Ethnicity/ Race	Total sample of dry adult sacra	Complete dorsal wall aggenesis	
				No	(%)
1	Trotter M <sup>[17]</sup>	USA	553	10	1.8
2	Kumar et al. <sup>[18]</sup>	India	202	3	1.49
3	Sekiguchi et al. <sup>[19]</sup>	Japan	92	1	1
4	Nagar SK <sup>[20]</sup>	India	263	4	1.5
5	Senoglou et al. <sup>[21]</sup>	Turkey	96	2	2.08
6	Patel et al. <sup>[22]</sup>	India	150	4	2
7	Kiran et al. <sup>[23]</sup>	India	50	1	2
8	Patil et al. <sup>[24]</sup>	India	103	3	2.91
9	Singh R <sup>[4]</sup>	India	140	2	1.4
10	Seema et al. <sup>[25]</sup>	India	159	5	3.14
11	Suwanlikhid et al. <sup>[26]</sup>	Thailand	235	1	0.43
12	Shewale et al. <sup>[27]</sup>	India	204	2	0.98
13	Kubavat et al. <sup>[28]</sup>	India	302	5	1.65
14	Ukoha et al. <sup>[29]</sup>	Nigeria	83	1	1.2
15	Nasr et al. <sup>[30]</sup>	Egypt	150	4	2.66
16	Kamal et al. <sup>[31]</sup>	Bangladesh	172	1	0.6
17	Nagendrappa RB <sup>[32]</sup>	India	100	3	3
18	Shinde V <sup>[33]</sup>	India	100	1	1
19	Mishra et al. <sup>[34]</sup>	India	93	4	4.3
20	Chhabra N <sup>[35]</sup>	India	32	1	3.12
21	Malarvani et al. <sup>[36]</sup>	Nepal	100	3	3
22	Akhtar et al. <sup>[37]</sup>	India	116	3	2.58
23	Saha D <sup>[38]</sup>	India	125	2	1.6
24	Shinde et al. <sup>[39]</sup>	India	300	4	1.33
25	Dhuria et al. <sup>[40]</sup>	India	88	3	3.4



No	First author	Ethnicity/ Race	Total sample of dry adult sacra	Complete dorsal wall agenesis	
				No	(%)
26	Aragao et al. <sup>[41]</sup>	Brazil	45	2	4.44
27	Bagoji et al. <sup>[42]</sup>	India	138	4	2.89
28	Pandey M <sup>[43]</sup>	India	86	2	2.33
29	Poudel et al. <sup>[44]</sup>	India	70	3	4.28
30	Yonkuc et al. <sup>[45]</sup>	Turkey	110	4	3.63
31	Punja et al. <sup>[46]</sup>	India	50	1	2
32	Abera et al. <sup>[47]</sup>	Ethiopia	61	1	1.63
33	Naznin et al. <sup>[48]</sup>	Bangladesh	60	3	5
34	Chandan et al. <sup>[49]</sup>	India	276	3	1.08
35	<b>Present study</b>	<b>Greece</b>	<b>155</b>	<b>3</b>	<b>1.93</b>
TOTAL			5059	99	



**Figure 4.** Incidence of complete dorsal wall agenesis of the sacral canal in different population groups.

## Medical implications

In theory, since the spinal cord terminates at around L2, spina bifida shouldn't cause any serious medical problems. However, there is evidence that it can affect various systems with serious consequences on medical procedures. The detailed knowledge of sacral anatomical divergences is of paramount significance for several medical specialists (orthopaedic surgeons, neurosurgeons, neurologists, urologists, anesthesiologists, obstetricians, radiologists).

## In orthopedics and neurosurgery

The association between SSBO and low back pain is obscure, but obviously the compression of the spinal nerves' roots that pass through the exposed sacral canal can be related to atypical low back pain (LBP).<sup>[4]</sup> A single study by Taskaynatan et al. suggested that 4.3% of the patients with low back pain were diagnosed with SSBO.<sup>[51]</sup> A possible mechanism of LBP in cases of complete DAS, is that the modification of the back muscles' attachment on the sacrum (e.g. erector spinae, multifidus) changes the biomechanical dynamics of the vertebral column.<sup>[4]</sup> In addition, due to the anatomical alterations of the area, SSBO patients often present with secondary pathological conditions of the spine, such as posterior disc herniation. Supposedly, the congenital defect could cause instability and lead to degenerative deformities and LBP.<sup>[52]</sup> Paraskevas et al. described a case of a dried sacrum presenting a partially sacralized fifth lumbar vertebra and total spina bifida, extended from first to fifth sacral vertebra.<sup>[53]</sup> Moreover, even minor external trauma is likely to cause fracture of an impaired sacrum with total SSBO. That probably explains the cases of sacral fatigue fractures in children with SSBO reported in the literature.<sup>[54]</sup> Presence of spina bifida occulta with a completely open SC increases the chances of iatrogenic injury of the sacral nerves during internal ilio-sacral screw fixation, which involves the fixation of the screws on the pedicle of S1 and S2 vertebrae. The correct surgical planning certainly poses a challenge for the orthopedic surgeon and neurosurgeon, since complications include neurological deficits, implant failure, and the need for second surgery.<sup>[55]</sup>

## In anesthesiology (CEB)

Caudal epidural block has been extensively used for the diagnosis and treatment of lumbar spinal disorders, for the management of chronic back pain and for the analgesia and anesthesia in labor and genitourinary surgery. For optimal access into the sacral epidural space, the apex of sacral hiatus and the sacral cornua are used as anatomical landmarks.<sup>[56]</sup> The absence of these landmarks, such as in DAS, may complicate the spinal anesthesia. Ultrasound is a safe, simple, and non-invasive method of preoperative examination of the sacral anatomy and increases the success rate of caudal epidural block by 100%.<sup>[57]</sup>

## In urology

SSBO, especially total SSBO, is suggested to be linked to a variety of functional disorders of the lower urinary tract. In 1985, Galloway and Tainsh found an increased number of spina bifida occulta cases in a small group of adults with lower urinary tract problems.<sup>[58]</sup> Some years later, Fidas et al. concluded that neurophysiological abnormalities in patients with dysfunction of the lower urinary tract may be associated with congenital dysraphic lesions in the lower lumbar spine and the sacrum. There appears to be no direct causal relation between the radiological and neurophysiological abnormalities, but the findings suggest a common etiology.<sup>[59]</sup> Reported bladder and urethral abnormalities in SSBO include detrusor hyperreflexia during filling, low bladder compliance, impaired bladder sensation, detrusor/sphincter dyssynergia, absent anal reflex. Wu et al. suggested that SSBO could gradually lead to dysfunction of spinal nerves and lower urinary tract symptoms, such as an overactive bladder.<sup>[50]</sup>

## Limitations of our study

Our sample size was rather small for safe and reliable documentation of the incidence of DAS in the Hellenic population and its relation to male sex. We suggest that further research should include greater numbers of Hellenic bone specimens. Sex as a possible etiologic or causative factor should be equally investigated.

## CONCLUSION

The dorsal wall of the sacrum presents with an abundance of anatomical divergences. The inter-population incidence of complete agenesis of the sacral canal's posterior wall ranges from 0.43% to 5% in the international literature. We found its incidence in the Hellenic population to be 1.93%.

The authors suggest that the future studies in different populations must mention the sex of the specimen, to determine it as a factor. More clinical studies on complete dorsal wall agenesis of the sacral canal are needed to establish pathophysiologic and genetic mechanisms. Awareness of anatomical variations is the key to successful results in the clinical setting; the complete agenesis of the SC dorsal wall (total spina bifida occulta) is not an exception to this rule.

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## Conflict of Interest

None.

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# Полная агенезия дорсальной стенки крестцового канала среди греческого населения: остеологический анализ

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## Резюме

**Введение:** Несостоятельность закрытия дорсальной стенки крестцового канала (КК) известна с самого начала современной остеологии, появившись ещё в доисторические времена. Варианты включают частичное или полное отсутствие дорсальной стенки КК. КК представляет собой путь для минимально инвазивных терапевтических и диагностических процедур при заболеваниях позвоночника и для обеспечения анальгезии и анестезии при операциях, включая роды и мочеполовую хирургию.

**Цель:** Наша цель – проверить частоту полной агенезии дорсальной стенки КК среди греческого населения.

**Материал и методы:** Мы собрали 155 взрослых сухих крестцов определённого пола с Третьего кладбища Афин для изучения дорсальной стенки крестца. Повреждённые или изменённые крестцы были исключены. Мы также провели исчерпывающий обзор соответствующей литературы и сравнили наши результаты с результатами международных исследований.

**Результаты:** Полная агенезия дорсальной стенки крестцового канала была обнаружена в трёх случаях (1.93%), у двух мужчин (1.29%) и одной женщины (0.64%). В нашем обзоре рассматривается его частота в других популяциях и изучается возможность региональной или расовой корреляции.

**Заключение:** Знание полной агенезии дорсальной стенки крестцового канала медицинскими специалистами имеет решающее значение для предотвращения осложнений при хирургии позвоночника, анестезии и акушерстве, а также при дифференциальной диагностике неврологических и урологических заболеваний.

## Ключевые слова

полная агенезия, греческий, крестец, вариации крестца, крестцовый канал, spina bifida