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Sacral Spina Bifida Occulta: A Frequency Analysis of Secular Change

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Abstract: Substantial relaxation of natural selection beginning around 1900 changed the mutation/selection balance of modern genetic material, producing an increase in variable anatomical structures. While multiple structures have been affected, the temporal increase in variations of the sacrum, specifically, 'Sacral Spina Bifida Occulta,' have been reliably demonstrated on a localised scale. Calculation of largescale frequency has been hindered by the localised nature of these publications, the morphological variability of this variation, and potential pathological associations, which have produced divergent classifications, and conflicting reported rates of occurrence. A systematic review of the reported literature was conducted to provide an objective analysis of Sacral Spina Bifida Occulta frequency from 2500 BCE to the present. This review was designed to compensate for observed inconsistencies in reporting and to ascertain, for the first time, the temporal trajectory of this secular trend. A systematic review of Sacral Spina Bifida Occulta literature was conducted through the strict use of clinical meta-analysis criteria. Publications were retrieved from four databases: PubMed, Embase, the Adelaide University Library database, and Google Scholar. Data were separated into three historical groups, (1 = <1900, 2 = 1900 to 1980 and 3 = >1980), and frequency outcomes compared, to determine temporal rates of occurrence.

A total of 39/409 publications were included in the final analysis, representing data for 16,167 sacra, spanning a period of 4,500 years. Statistically significant results were obtained, with total open S1 frequency increasing from 2.34%, (79 to 1900CE), to 4.80%, (1900 to 1980CE) and to 5.43% (>1980CE). These increases were significant at $p<0.0001$, with Chi-squared analysis. A clear secular increase in the global frequency of Sacral Spina Bifida Occulta has been demonstrated from 1900 to the present. This research provides a novel and adaptable framework for the future assessment of variation distribution, with important implications for the fields of biological anthropology and bioarchaeology.

Key words: Sacral Spina Bifida Occulta (SSBO), frequency, classification, natural selection
Abbreviations: Sacral Spina Bifida Occulta (SSBO), Spina Bifida Cystica (SBC), Neural Tube Defect (NTD)

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Introduction

The relaxation of natural selection can be attributed to the decreased rate of infant mortality and the increased rate of adult survivability from 1900 onwards (Ulizzi et al. 1998). These changes were shaped by improved clinical understanding of disease, the invention of increasingly effective medication, and an improvement in prenatal and postnatal medical care (Ruhli and Henneberg 2013; Solomon et al. 2009). Consequently, survivorship to the age of reproduction (15 years) increased from <50% in 1850 to slightly >90% by 1900 (Greene 2001). Overall, the probability that an average person born into a population will be able to pass their genes to the next generation rose from 0.30 to 0.95 (Saniotis and Henneberg 2011). This reduction in the opportunity for selection, altered the mutation/selection balance which precipitated phenotypic variation (Cairnes and Gariepy 1990; Lee et al. 2011). Such an increase has been observed in a number of modern physiological, immunological, and morphological characteristics, the most well-known of which is the increase in lactose intolerance and the congenital absence of the third molar (Ingram et al. 2009; Swallow 2003). Occurring over a relatively short period of evolutionary history, (120 years), these modern secular changes have been observed in multiple correlating anatomical structures.

One such example includes increases observed in the retention of the embryonic variant, the median artery. This embryonic vessel typically regresses at 8 weeks gestation, but retention of this artery into adulthood has experienced an increase of 20%, over a period of the last 170 years (Lucas et al. 2020). The atypical fusion of one or more tarsal bones of the foot has also been subject to observed increases after 1900, with an increase of >12%, evidenced over a period of 50 years (Ruhli et al. 2003). The timing of these changes in anatomical structures, coincides with observed increases in sacral variations, most specifically the 'vertebral anomaly' Sacral Spina Bifida Occulta, (SSBO).

Sacral Spina Bifida Occulta (SSBO) is a condition difficult to define due to the variability with which it is described in the literature, and the broad spectrum of defects this condition can represent (Albrecht et al. 2007; Eubanks and Cheruvu 2009). SSBO is often considered the mildest manifestation of Neural Tube Defect (NTD), specifically of the highly debilitating Spina Bifida Cystica (SBC), which has been identified as the most common congenital anomaly of the 21st century (Kallen and Lofkvist 1984; Morrison et al. 1998). Characterised skeletally, by the absence or non-fusion of one or multiple posterior vertebral arches, SSBO variably includes deformation of the laminae, neural arch, or pedicles of vertebrae (Post 1966; Sutow and Pryde 1955). While this anomaly can occur at any level of the vertebral column, the malformation of the last lumbar vertebra and the first sacral vertebra is the most routinely observed, studied, and reported (Sairyo et al. 2006). Due to the severity of deformation caused by SSBO, (which typically presents as the exposure of the sacral canal, or absence of the dorsal wall), this condition is easily identifiable in dry human sacra, and can be reliably distinguished from post-depositional erosion or damage, (Figs 1, 2 and 3). Therefore, observations of this condition in dry human sacra are reliable, and publications which provide

frequency data in this context can be assumed to be accurate and objective.

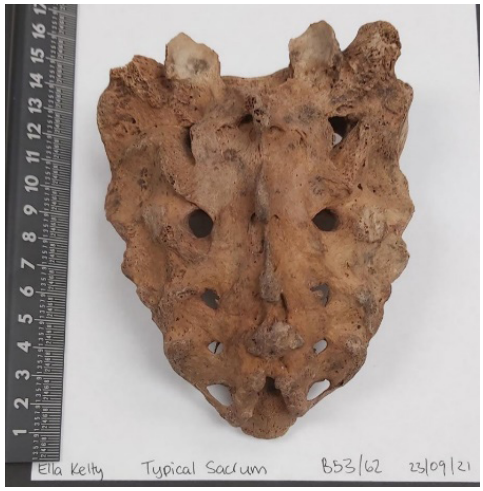

Fig. 1. Dry human sacrum with a typically formed dorsal wall – fully fused sacral vertebrae. (Photograph taken by lead author (Kelty 23/09/2021). Specimen B53 from St Marys archaeological collection, ethically held by The University of Adelaide).

Fig. 2. Dry human sacrum demonstrating 'Total SSBO' or non fusion of arches of all sacral segments. (Photograph taken by lead author (Kelty 23/09/2021). Specimen B79 from St Marys archaeological collection, ethically held by The University of Adelaide).

Fig. 3. Dry human sacrum demonstrating non-fusion of sacral segments, S1 and S4-S5. The most commonly observed configuration of S1 non-fusion. (Photograph taken by lead author (Kelty 23/09/2021). Specimen B61 from St Marys archaeological collection, ethically held by the University of Adelaide).

Deformation at all levels of the sacrum can occur with varying degrees of regularity, dependent on the sacral segment involved. The most common observation of non-fusion occurs at segments S4-S5, which can reach upwards of 90% of individuals with European ancestry (Fidas et al. 1987). Thus, this deformation pattern is clinically recognised as a natural morphological variation, termed the sacral hiatus (Abera et al. 2021; Henneberg and Henneberg 1999). Deformations of segments S2 and S3 have lower frequencies, (1% to 10%), but to date are also considered natural variations, due to their sometimes inclusion into the hiatus (Simriti et al. 2017). This inclusion of S2-S5 in the natural variation of the sacrum suggests that

these specific patterns of deformation have no pathological associations and are therefore of no clinical importance (Kumar and Tubbs 2011).

Non-fusion of the first sacral segment usually has a lower frequency, similar to that of S2 and S3, but is unreliably reported, with estimations ranging from 8% (Piontek 1971) to 23% (Sairyo et al. 2006). Unlike the segments S2-S5, S1 has clear pathological associations having been reliably correlated with enuresis, posterior disk herniation, and lower back pain (Eubanks and Cheruvu 2009; Sutow and Pryde 1955). Non-fusion of sacral segments inclusive of S1 can be considered morphologically and clinically important, irrespective of non-fusion or fusion of other segments. It is for this reason that Sacral Spina Bifida Occulta can be specifically defined as non-fusion inclusive of the first sacral segment (Henneberg and Henneberg 1999; Lee et al. 2011; Solomon et al. 2009).

The Pelvis (Os Coxae) is the most variable aspect of the human skeleton due to its high levels of sexual dimorphism, with the sacrum being considered the most variable aspect within that structure (Steyn and Iscan 2008). Variations to the structure of the sacral canal can also influence variation in the resulting morphology of the surrounding Os Coxae (Kurki 2013). The degree to which these variations can influence pregnancy, birth, overall health, and forensic sex identification, has resulted in a relative wealth of clinical, anthropological, and archaeological assessments of this variation over the last century (Henneberg and Henneberg 1999). The identification and classification of Sacral Spina Bifida Occulta was first described in the anthropological literature by Willis (1923). Willis popularised the characterisation of this condition as a 'vertebral anomaly' of no clinical significance, recording only a 1.2% frequency in 748 historical subjects (Willis 1923). Anthropological interest in SSBO was shaped by this definition but was characterised by inconsistency in reported frequencies. Ferembach (1963) famously reported a 76% frequency in a sample from 12,500 BCE, but this was hard to substantiate, due to the small sample size and the 8% to 23% occurrence which typified the literature of this period. Inconsistencies in reported SSBO frequencies were further exasperated by the clinical recognition of this condition, which aligned with investigations of neural tube defects in 1980 and introduced a new generation of conflicting classifications and frequency calculation methods (Molloy et al. 2017; Scatliff et al. 2013).

Investigations into the temporal increase of SSBO frequency in the modern era, and its correlation to the relaxation of natural selection around 1900, have ultimately been impeded by the number of academic debates, controversies and disagreements which characterise this research area (Shore 1930; Zemirline et al. 2013). A long-standing consensus within the medical community that anatomically modern humans are no longer evolving under the operation of natural selection, has prevented largescale research into these changing anatomical structures and their potential impact on the health of future populations (Kumar and Singh 2003; Rühli and Henneberg 2013). While small scale and localised studies have been conducted which reliably support this correlation between various changing modern anatomical structures and the relaxation of natural selection in the industrialised world from 1900 onwards, (Lucas et al. 2020; Rühli et al. 2003) large-scale assessments and

widespread acceptance of this phenomenon have yet to be established.

This observed lack of academic consensus has prevented reliable calculation of SSBO frequency over time, which is additionally impeded by the small number of publications which contain reliable data for this condition (Zemirline et al. 2013). These inherent limitations have been addressed by modern SSBO research, which provides more reliable assessments of frequency than historically observed (Kumar and Singh 2003). Interestingly, an 11% frequency of this condition was observed in Pompeii (79CE, Henneberg and Henneberg 1999), being about one half of modern European assessments of about 20% (Saluja 1988). This led Henneberg and Henneberg (1999) to suggest that a secular and microevolutionary trend could be observed in SSBO frequency. This increase was further substantiated by Solomon et al. (2009) and Lee et al. (2011), who demonstrated an increase in the frequency of SSBO at S1 in Australian and European birth cohorts, from 1940s to 1980s. This is interesting, as these localised studies demonstrate an increase that not only correlates with the observed relaxation of natural selection around 1900, but that also coincides with relative increases in similar anatomical variations during the same period. It is therefore hypothesised that the generation of a large-scale, geographical, and temporal assessment of SSBO frequency will produce evidence of a clear secular trend in the increase of this condition from 1900 onwards.

Materials and Methods

A literature review was performed to collect all available publications pertaining to SSBO frequency as previously defined. This review generated a total of 409 foundational or peer-reviewed publications. Predetermined exclusion criteria were used to determine the relevance of each publication and assess the quality of their reported segmentation data (Fig. 4). In total 39 of 409 (<10%) publications were included in the frequency analysis, producing a total sample size of 16,167 sacra, which spanned 25 international regions (Fig. 5) and a period of 4,500 years. Male and female sample sizes were also recorded where reported, with a total male sample size of 3,992 and female sample size of 3,818, with 8,357 (51.69% of 16,167) having undesignated sex.

In order to reliably evaluate the true frequency of SSBO it was necessary to design a method that could enable the review of all available and relevant literature, while overcoming observed inconsistencies in classification and frequency calculation. It was also imperative to demonstrate that modern human skeletal anatomy is subject to evolutionary change, and that increases in SSBO frequency directly correlate with the recent relaxation of selection shift. The literature was collected, assessed, and analysed according to strict clinical meta-analysis guidelines to ensure that data were reviewed systematically (Balduzzi et al. 2019; Page et al. 2021). As this research does not contain clinical trials or patients, some meta-analysis criteria could not be applied, and the decision was made to conduct a quantitative literature review/frequency analysis instead. To guarantee cohesion, validity and accuracy within the research design, all analyses were conducted according to the requirements of a meta-analysis where possible (Higgins et al. 2003; Page and Moher 2017).

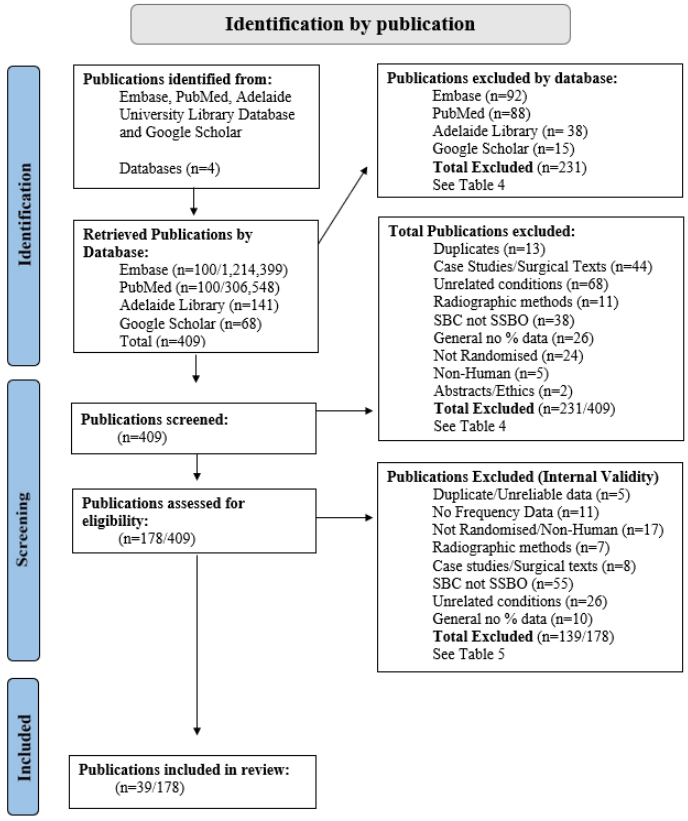

Fig. 4. PRISMA Flow chart of literature analysis method, with added inclusion/exclusion criteria.

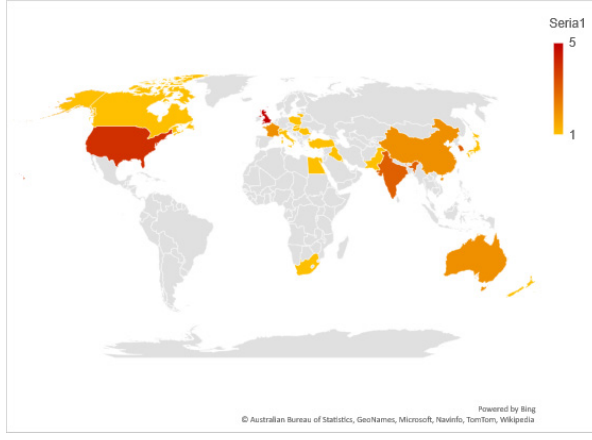

Fig. 5. World map showing distribution of included SSBO data for the literature analysis. Country of origin and number of publications per country included.

Literature Review Method

Publications for the literature review were collected through the use of Embase, PubMed, Google Scholar, and the University of Adelaide's library database. Embase and PubMed were used primarily for the collection of clinical literature relating to SSBO and did not include anthropological and archaeological data for this condition, particularly not from the early 20th century. Google Scholar was used as a means to find those publications not available in the medical literature databases, and the University of Adelaide Library was used to gain access to those publications found in Google Scholar that were restricted by paywalls. The Adelaide University Library database was the most practical resource to use for supplementary access to these publications, as both authors are members of The School of Biomedicine at this University.

Databases were searched for keywords; *Sacral Spina Bifida Occulta, Spina Bifida Occulta, Neural Tube Defect, Spina Bifida* and *Occult Spinal Dysraphism*. Keywords were supplemented by corresponding searches for, *incidence, prevalence, frequency,* and *rates*. Results from each database underwent two rounds of screening, the initial publication screening, (12/2/2021 – 03/05/2021) and the internal validity screening, (13/09/2021 – 2/10/2021), which included different criteria. The initial publication screening was conducted through a process of examining the abstract, results and conclusions of each publication, and including/excluding each publication based on a set of predetermined exclusion criteria (Fig. 4) (Balduzzi et al. 2019). Publications that were included through the initial publication screening were then analysed a second time with more stringent predetermined exclusion criteria, which focussed on the assessment of the internal validity of each study (Page et al. 2021).

Different retrieval strategies were used dependent on the database. PubMed and Embase are clinical databases that were used to source potential clinical data on the frequency of SSBO. Due to the volume of publications generated from such expansive databases, as a result of the search strategies outlined (Table 1), only the top 100 search results were included for screening. The University of Adelaide library and Google scholar databases were used primarily to retrieve anthropological and archaeological data on the frequency of this condition. Due to the nature of these databases, specific search strategies were not used, however, each afore mentioned key word was searched, and any relevant publications were retrieved. This was further complemented by the use of these databases to retrieve publications cited in already analysed works and to expand upon the key words to include, *sacral hiatus, paleoepidemiology, sacral anomaly, sacral deformity and osteoarchaeology*.

It should be noted that due to the nature of SSBO and its presence in historical and archaeological populations, this review of the literature is amalgamating already published data on the frequency of this condition in dry human sacra, from cadaver studies and through anonymised radiographic data. Therefore, this review is bio-anthropological and does not include patients, clinical trials, medical equipment, additional reviewers, ethics approval or funding grants. All publications were reviewed by the lead author manually, no automation process or equipment was used, and no additional reviewers or external parties were involved.

Table 1. Search strategies for publication retrieval from each database

Database	Keywords	Search String*	Retrieved/ Generated*
PubMed	Sacral Spina Bifida Occulta, Spina Bifida Occulta, Neural Tube Defect, Spina Bifida and Occult Spinal Dysraphism. + Incidence, Prevalence, Frequency, and Rates	(Sacral Spina Bifida Occulta) OR (Spina Bifida Occulta) OR (Neural Tube Defects) OR (Spina Bifida) OR (Occult Spinal Dysraphism) AND (Incidence) OR (Rates) OR (Frequency) OR (Prevalence)	100/ 306,548
Embase	Sacral Spina Bifida Occulta, Spina Bifida Occulta, Neural Tube Defect, Spina Bifida and Occult Spinal Dysraphism. + Incidence, Prevalence, Frequency, and Rates	Exp spinal dysraphism / or exp neural tube defects / or exp open spinal dysraphism / and exp incidence / or exp frequency / or exp prevalence /	100/ 1,214,399
The University of Adelaide	Sacral Spina Bifida Occulta, Spina Bifida Occulta, Neural Tube Defect, Spina Bifida and Occult Spinal Dysraphism. + Incidence, Prevalence, Frequency, and Rates + Sacral Hiatus, Paleoepidemiology, Sacral Anomaly, Sacral Deformity and Osteoarchaeology	N/A	141
Google Scholar	Sacral Spina Bifida Occulta, Spina Bifida Occulta, Neural Tube Defect, Spina Bifida and Occult Spinal Dysraphism. + Incidence, Prevalence, Frequency, and Rates + Sacral Hiatus, Paleoepidemiology, Sacral Anomaly, Sacral Deformity and Osteoarchaeology	N/A	68

*Search string and number of publications generated are not applicable to the University of Adelaide Library and Google scholar databases.

Initial publication screening parameters

Once all 409 publications had been retrieved, they underwent the initial screening process and were included/excluded based on stringent predetermined criteria, (Fig. 4). As the primary objective of this literature analysis was to obtain frequency data for SSBO, publications that did not include frequency data for this condition, data for SSBO specifically, or those that did not include random samples, were excluded. Case studies which discussed only single examples and therefore had no frequency data were excluded. Surgical texts were also excluded on the basis that they related to the diagnosis, management, treatment, and

surgical outcomes of spinal dysraphism. As such the frequency of occurrence was not reported, as all individuals observed had already been diagnosed with this condition. A singular list of abstracts for a conference on the neurosurgical management of spinal dysraphism was also excluded due to lack of detailed frequency data. Publications detailing novel radiographic methods for the identification of SSBO were also excluded, as prevalence data were not reported. One publication pertained solely to ethics, one was in reference to widescale arsenic poisoning, and a number were related to non-human clinical trials which were of no relevance to this research.

'General SSBO' included publications that were designed as informative documents on the identification, diagnosis, and treatment of SSBO from a clinical perspective. These publications did not include frequency data, and more than half were in reference to SBC not SSBO. This misidentification of SSBO as the neural tube defect SBC, was also an exclusion criterion. A number of publications retrieved from Embase, and PubMed also included publications on pathologies completely unrelated to SSBO. Issues in identifying SSBO data specifically, were further complicated by the number of associated pathologies researched clinically. Publications relating to these pathologies, were also assessed, and included only if the frequency data were wholly separated from those of the associated pathology, and if adequate and appropriate control groups were used (Page and Moher 2017; Page et al. 2021).

A disproportionate number of the retrieved publications were related to Spina Bifida Cystica (SBC) and Neural Tube Defects, and included no reference to, or data for SSBO. This was the consequence of an early proposal to compare frequency data for these two conditions to ascertain the importance of their relative patterns of occurrence. As this research progressed, it was determined that SBC frequency was already reliably established in the literature, and therefore these studies were not included in the final analysis (Fig. 4). Trusted data for SBC, however, were obtained from national and global birth registers, derived from these excluded publications, to compare the relative prevalence of this condition with that of SSBO for the same period (Atta et al. 2016). This was achieved through the calculation of mean values for reported births with SBC per 1000, in European populations. These are not included in the results but were generated for the purpose of aiding the discussion.

Internal Validity Screening Parameters

Once these publications had been screened for the more basic parameters, (inclusion of SSBO frequency data), the remaining 178 publications were subjected to an additional, more stringent, screening process, to access the internal validity and address the risk of bias in their results (Higgins et al. 2003). The quality of included segmentation data was assessed, and those that did not include data for deformation of S1 specifically, or which reported duplicate data, were excluded. Archaeological and anthropological texts from the early 20th century which included purely textual anecdotes, were thoroughly scrutinised to ensure that sample sizes and case numbers were accurate and did not contain any missing or unclear data. Any uncertainty as to the clarity, totality, or accuracy of the data

from these publications resulted in them being excluded to ensure the generation of meaningful and reliable results (Higgins et al. 2003). Publications that included duplicate data already screened in previous publications, or data that could not be reliably differentiated from other osteological assessments from similar or sometimes the same archaeological sites, were also excluded.

Those publications which assessed the association between SSBO, and a range of pathologies were also assessed to ensure that frequency data for SSBO were wholly separable from those of the associated pathology, and that adequate control groups had been used. Those which did not provide adequate control groups, or studies which included only patients with a pathology, or deformity, reliably associated with SSBO, (eg: Cutaneous stigmata), were excluded on the basis that they did not represent the true frequency of this condition. Radiological assessments of this condition, which focussed on novel methods for the identification of SSBO, were also excluded if they contained zero or duplicate frequency data for this condition, or if the level of deformity, (segment), was not reported.

Once completed, this review of the current literature produced 39/409 publications for inclusion into the frequency data analysis. The included publications, as outlined in detail in the appendix, ranged in publication date from 1932 to 2019, 30 of these were peer reviewed, with the remaining nine having been published before the introduction of the peer review system. All included publications reported SSBO frequency data that were analysed and deemed reliable, and no publication was assessed which examined the frequency of SSBO and did not produce at least one case of this condition.

Frequency Analysis Methods

A total of 39/409 publications were included in the final frequency analysis having conformed to the outlined inclusion criteria, (Fig. 4). Numbers were allocated to each publication and citation, location and dating details were recorded for each. Reported case numbers of identified SSBO were divided by reported sample sizes, and multiplied by 100, to produce percentage values. This was completed for each possible combination of reported deformation, across all sacral segments. This included deformation of segments inclusive of S1, (ie: L5-S1, S1andS5), and calculation of male and female frequencies (Henneberg and Henneberg 1999; Lee et al. 2011; Solomon et al. 2009).

Recalculations were made where reported prevalence was not clearly presented, with some cases and sample sizes being combined where necessary, (control/patient and multiple juvenile samples). Patient groups that were proven to be random (not commonly or primarily associated with SSBO) were combined with control group sample sizes, and case numbers, to determine frequency for the whole group. Publications that separated subadults (1–15 years) into smaller sub-divisions of age, (eg: 1–2 years, 3–4 years etc.) were also grouped together, and an identical method was used to determine the relative frequency (Page et al. 2021). Similar additions were also made with the male and female frequency calculations. This occurred where male and female cases were recorded for both the control and patient groups, which were then combined to determine the frequency, as per the method outlined above. Instances where sex was separated into age categories, of girl/boy, female/male structure, were also combined to determine the frequency by sex (Fidas et al. 1987).

Once this information had been collected for all 39 publications, the resulting data were separated into three distinct historical groups. This was done to consider the 4,500-year time span, to test the hypothesised increase of this condition after 1900 and 1980 and to ensure that each study would be accurately weighted. This separation was determined according to calculated date of birth of each group. Birth dates were either used as reported in more modern publications or estimated by subtracting average life expectancy figures from burial dates for historical collections (WHO 2012; WHO 2020). Historical Group 1 (HG1) included date range 2,500BCE to 1,900CE, the second Historical Group (HG2) encompassed all material dating from 1,900 to 1980CE while Historical Group 3 (HG3) included the remaining data for the period 1980CE to 2020CE. Male and female frequency data, where available, were also separated into historical groups, although an absence of reported sex data for HG3 did affect the results of this group.

Statistical methods

While data for SSBO were recorded for each sacral segment, only data for deformation inclusive of S1 were included into the statistical analysis. Total sacra observed, and total number of cases were determined for each historical group. Contingency tables were generated in the SSPS.25 software, (Tables 2 and 3) and Chi-squared calculation with Yates's correction, and corresponding *p*-values, were used to assess the direction of effect for these three groups (Henneberg and Henneberg 1999; Lee et al. 2011; Solomon et al. 2009). The available male and female data for all three historical groups were treated in the same way.

All statistical calculations were performed using the SSPS.25 software by the primary author with instruction and assistance from the secondary author. No external resources were used to complete this analysis and no additional reviewers were integrated into the assessment.

Table 2. Contingency table used to generate Chi-Squared statistic for the total frequency of SSBO

	Historical Group 1 <1900	Historical Group 2 1900-1980	Historical Group 3 >1980
Total Sacra Observed	6,901	8,074	1,192
Total SSBO Cases Identified	922	1,503	281

Total frequencies determined for comparison of Historical Groups 1 and 2, 2 and 3 and 1 and 3.

Table 3. Contingency table used to generate Chi-Squared statistic for the male/female frequencies of SSBO

	Historical Group 1 <1900	Historical Group 2 1900-1980	Historical Group 3 >1980
Total Male Sacra Observed	790	2,883	319
Total Male Cases of SSBO	98	738	46
Total Female Sacra Observed	720	2,830	268
Total Female Cases of SSBO	69	458	74

Male and Female frequencies determined through comparison of Historical Groups 1 and 2, 2 and 3 and 1 and 3.

Results

A total of 39 publications were included from the 409 retrieved and screened during the literature review process (Fig. 4). The results of both screening processes, with exclusion/inclusion criteria outlined for each publication, throughout each process, are presented in the appendix. Citation details and exact frequency data collected from each included study are also included in the appendix.

The frequency analysis results demonstrated a clear and statistically significant increase in the frequency of SSBO after 1900 (Table 4 and Fig. 6). The calculation of the total frequencies for historical groups one and two, evidenced a 5.25% increase in SSBO frequency, (<1900 to 1980), Chi-squared 54.503 (p<0.0001). The comparison of total frequencies between historical groups two and three also provided a very statistically significant result, with a 4.98% increase, Chi-squared 10.543 (p<and a p-value of <0.0012). Total frequency comparison was also completed between historical groups one and three, which demonstrated an increase of 10.23% from the period <1900, to the present, Chi-squared 57.843 (p<0.0001).

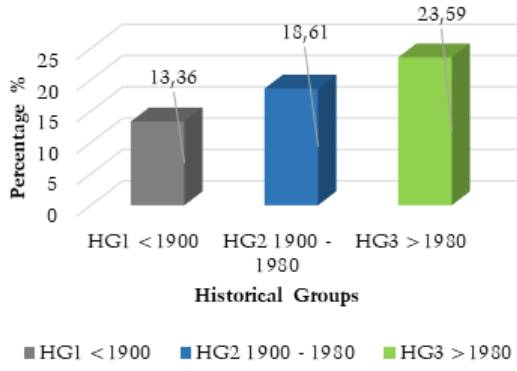

Fig. 6. Total SSBO frequency by historical group. Data derived from numerical analysis of included literature and frequency calculations outlined in Table 4.

Table 4. Total data included in each historical group

	Historical Group 1 <1900	Historical Group 2 1900-1980	Historical Group 3 >1980
Total Included Publications	18	17	7
Total Included Studies	28	40	7
Total Sacra Observed	6,901	8,074	1,192
Total SSBO Cases Identified	922	1,503	281
Total Frequency	13.36%	18.61%	23.59%
Total Increase	2.40%	5.25%	4.98%
Chi-Square Value	HG1-HG2 54.503	HG2-HG3 10.543	HG1-HG3 57.843
p-Value	<0.0001%	<0.0012%	<0.0001%

Detailed representation of the total data for each historical group includes significance calculations and data used to calculate frequency.

The male and female frequency calculations for the three historical groups also produced statistically significant results for an increase of SSBO after 1900 and 1980. A clear increase was demonstrated in males (13.19%) and females (6.60%) for historical groups one and two, generating Chi-squared values of 40.618 and 14.737 (for both $p<0.0001$). A decrease was observed in male frequency of -11.17%, (Chi-squared=12.209, $p<0.0005$) between historical groups two and three, but an increase was demonstrated for females (Chi-squared = 14.105, $p<0.0002$). (Table 5 and Fig. 7).

Table 5. Male and female data included by historical group

	Historical Group 1 <1900	Historical Group 2 1900-1980	Historical Group 3 >1980
Total Included Publications	6	10	4
Total Included Studies	10	23	4
Total Male Sacra Observed	790	2,883	319
Total Males Cases of SSBO Identified	98	738	46
Total Frequency	12.40%	25.59%	14.42%
Total Increase/Decrease	N/A	+13.19%	-11.17%
Chi-Squared Value	N/A	HG1-HG2 40.618	HG2-HG3 12.209
P-Value	N/A	<0.0001%	<0.0005%
Total Female Sacra Observed	720	2,830	268
Total Female Cases of SSBO Identified	69	458	74
Total Frequency	9.58%	16.18%	27.61%
Total Increase/Decrease	N/A	6.60%	+11.43%
Chi-Squared Value	N/A	HG1-HG2 14.737	HG2-HG3 14.105
p-Value	N/A	<0.0001	<0.0002

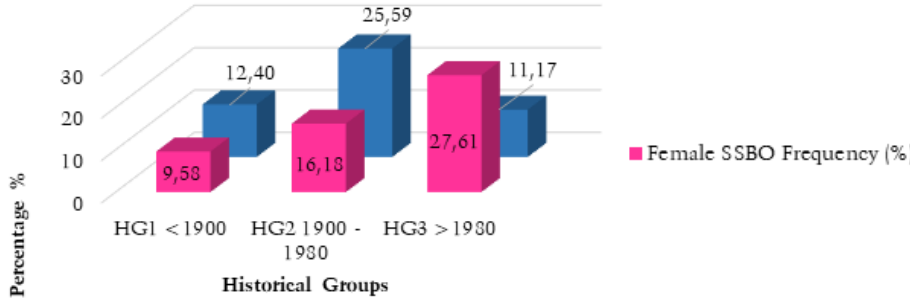

Fig. 7. Male and female frequency by historical group. Data derived from numerical analysis of included literature and frequency calculations outlined in Table 8.

Discussion

Almost all results pertaining to the overall frequency of SSBO demonstrated a substantial, statistically significant increase after 1900. These results provide compelling confirmation for the hypothesised increase in the frequency of SSBO and its correlation with the relaxation of natural selection around 1900. The importance of these results for the determination of current evolutionary change can be conceptualised by outlining the frequency for each historical group. HG1, (2,500BCE to 1900CE), had a total frequency of 13.36%, compared to HG2, (1900 to 1980) at 18.61%, which demonstrates a clear increase of 5.25% over a small 80-year period. HG3, (>1980) produced a frequency of 23.59%, and an increase of 4.98% from HG2, despite representing a smaller sample size and shorter period of history (40 years). When compared to HG1 (<1900), HG3 demonstrated an even more significant result, of a 10.23% increase in the frequency of SSBO between 1900CE and the present. When converted to increase per decade, these figures: 1.31% and 2.49%, demonstrate an accelerating non-linear increase in the frequency of SSBO in the modern era (Saniotis and Henneberg 2011).

The calculation of male and female prevalence for HG1 and HG2 also produced statistically significant results in support of an increase of SSBO after 1900. These results demonstrated higher percentages of increase than the total frequency calculation for these historical groups. Despite the sample sizes of each sex (3,992 males and 3,818 females) being similar, male prevalence was much higher and demonstrated a 6.59% greater increase than among females (13.19% male to 6.60% female) between HG1 and HG2. This could potentially represent a sex based evolutionary trend that has yet to be fully investigated, as male frequency was consistently higher with the exception of the decrease observed for males in HG3. This 11.17% decrease observed for males after 1980 is the only decrease noted across the entire analysis and is accompanied by a substantial 11.43% increase for females in this group. These calculations for >1980 are based on just four publications, with small numbers of sacra, and thus, may reflect regional differences rather than temporal trends. It is important to note that additional data are needed for HG3 to ensure that these relative increases/decreases can be substantiated.

While this research does represent the largest assessment of SSBO in the literature (16,167 sacra), the scarcity of relevant literature and the acknowledged limitations of these studies suggest that these results represent only a fraction of potentially recoverable data. The increase in the frequency of SSBO is relatively modest compared to the median artery, which shows an increase of 20% over the same 120-year period, about double that of the increase in SSBO (Lucas T et al. 2020). Similarly, tarsal coalitions experienced an increase of 12% over a 50-year period, double the reported frequency demonstrated for SSBO for this period (Ruhli et al. 2003). These modest increases in frequency recorded for SSBO may be the product of the lack of data and academic consensus. It may be possible that with the inclusion of additional, larger, and targeted datasets, rates of SSBO frequency may increase again, to parallel those observed in these other anatomical structures.

This research holds important implications for the general application of

both biological anthropology and bioarchaeology. The recognition of implications of natural selection on widescale secular change can improve the accuracy of differential diagnosis in skeletal remains. Awareness of this increase in skeletal variation, its frequency, and patterns of presentation, can improve future bioarchaeological interpretations of trauma, pathology, and health status. The potential for this phenomenon to produce previously unobserved skeletal changes must also be recognised and attempts to identify pathological associations with new forms of variation must be addressed. By acknowledging that the human skeleton is changing, collaboration with the medical community and the use of clinical methodology, can strengthen the capacity of bioarchaeology to provide insight into global future health outcomes as they relate to secular changes.

The incorporation of clinical parameters, statistical calculations, and bias assessments into this bioarchaeological assessment of SSBO frequency has provided a unique opportunity to design a systematic methodology which can be applied to a range of skeletal and anatomical variations. This framework has allowed for the traditionally small scale and localised anthropological datasets to be amalgamated into a broad temporal and geographic 'map' of SSBO frequency, emphasising overarching patterns not identifiable in smaller studies. This method allows for small datasets to be incorporated with a high degree of accuracy and can facilitate a continuous addition of new data. Potentially, this could produce an ever increasing 'map' of SSBO frequency, where the addition of datasets from a range of researchers would allow the eventual creation of a truly global representation of SSBO frequency and its secular trajectory. This method could then be expanded to include additional anatomical variations, from independent or future researchers, that would also lead to the creation of global 'maps' of diverse conditions frequencies.

Future bioarchaeological assessments of skeletal variation should be reconceptualised, with the traditional focus on individual and localised assessments of change replaced with wide reaching systematic evaluations of broad scale frequency. Clear patterns of secular change could be reliably assessed on a global scale, and these trends systematically compared. The potential for this style of analysis to identify trends that have explicit implications for public health and medicine, can be demonstrated through the comparison of SSBO and SBC frequency. The 4.98% increase in SSBO frequency observed after 1980 demonstrates a sustained increase of this condition and conforms with Solomon et al. (2009) and Lee et al. (2011) results on the confirmation of this microevolutionary increase and secular trend, despite the introduction of folate supplementation in 1980. This is in direct opposition to expected clinical outcomes for SSBO after folate supplementation introduction, which has resulted in a sharp decline of NTD related births worldwide after 1980, (Fig. 8) (Atta et al. 2016). This would suggest that SSBO potentially does not follow the same embryonic and etiological trajectory as SBC and has a separate cause altogether. While these results do not confirm or identify the underlying cause of SSBO, the large scale and systematic nature of this assessment, provides the foundation to test such hypotheses further.

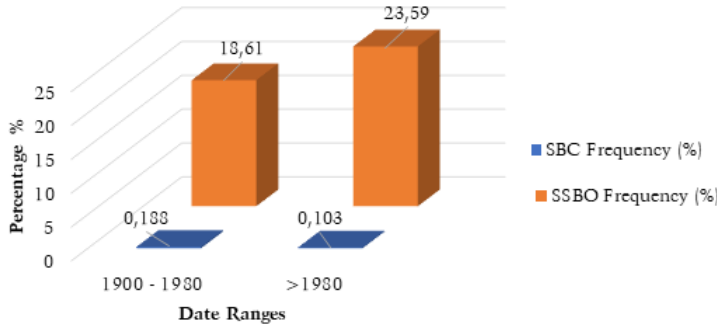

Fig. 8. Comparison between SBC and SSBO frequency by date. Resulting frequency data from literature analysis compared with reliably reported SBC prevalence.

While the results of this literature analysis were limited by the data available in the current literature, the primary objective of this analysis was achieved. The framework that this analysis has provided will facilitate the inclusion of additional SSBO data which will expand our understanding of this little-known condition and provide a uniform structure to ensure the replicability of all future research. In combination with the establishment of the most reliable frequency calculation to date, this framework will also enable the investigation of untested aspects of this condition, such as underlying etiology and additional pathological associations. Other anatomical variations, which have yet to be systematically evaluated, could also be incorporated into this framework, to establish a broader understanding of the trajectory and implications of secular evolutionary change in modern human populations.

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Authors' contribution

Both authors formulated the hypothesis. ERK collected data and drafted the text. MH helped with the analysis and edited text.

Conflict of interests

The Authors have no competing interests concerning this review.

Registration and Protocol

This review has not been registered as it is bioarchaeological.

The Protocol used for this review can be accessed in the appendix.

No amendments have been made due to registration or protocol.

Data Availability

PRISMA Flow Chart template 2020: http://www.prisma-statement.org/

Data for each individual study included in this analysis is available in the appendix.

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Supplementary Material

Table 6. Results of the initial screening process. Inclusion/exclusion criteria and number of publications included/excluded by database outlined

Exclusion reason	Number Excluded: Embase (n=)	Number Excluded: PubMed (n=)	Number Excluded: Adelaide Library (n=)	Number Excluded: Google Scholar (n=)	Total (n=) /409
Duplicates	9	2	1	1	13
Case Studies	11	5	1	1	17
Surgical Texts	12	13	2		27
Responses/Abstracts/Reviews	1				1
Radiographical Methods	6	3	2		11
Ethics		1			1
Non-Human Studies	1	3		1	5
General no % data	6	16	1	3	26
SBC not SSBO	10	13	10	5	38
Unrelated Clinical Conditions	26	25	16	2	68
Nonrandomised	11	7	4	2	24
Total Included	08/100	12/100	103/141	53/68	178
Total Excluded	92/100	88/100	38/141	15/68	231

Table 7. Results of the internal validity screening process. Inclusion/exclusion criteria and number of publications included/excluded by database outlined

Exclusion Criterion	Publications Excluded (n=)
Duplicate data	3
Unreliable data	2
Case Studies	5
Surgical Texts	3
SBC not SSBO	55
No segment data	5
No S1 data	6
Nonrandomised	15
Non-Human	2
Radiograph method	7
General no % data	10
Unrelated condition	26
Total Included	39/178
Total Excluded	139/178

Table 8. Included studies with sample size, date of samples as well as characteristics and any necessary assessments for bias in each publication

(n=)	Publication	Sample Size + Date	Characteristics	Risk of Bias Assessment*
1	Zemirline A et al. Lumbo-sacral malformations and spina bifida occulta in medieval skeletons from Brittany. Eur J Orthop Surg Traumatol. 2013;23: 149–153.	30 768 CE	Archaeological study of recovered skeletal human remains. (Dry human sacra)	Anecdotal data Clear, accurate and included segment data for SSBO, S1
2	Molto JE, et al. The paleoepidemiology of sacral spina bifida occulta in population samples from the Dakhleh Oasis, Egypt. Int J Palaeopathol. 2019;26: 93–103.	116 116 BCE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
3	Urrutia J, et al. Spondylolysis and spina bifida occulta in paediatric patients. Prevalence study using computed tomography as a screening method. Eur Spine J. 2016;25: 590–595.	228 2005 CE	Radiographic study of live patients with associated pathology.	CT and well-structured numerical results. SSBO and Spondylolysis data separate Clear, accurate and included segment data for SSBO, S1
4	Saluja PG. The incidence of spina bifida occulta in a historic and a modern London population. J Anat. 1988;158: 91–93.	112 1816 CE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
5	Lee YC et al. Confirmation of microevolutionary increase of spina bifida occulta among Swiss birth cohorts. Eur Spine J. 2011;20: 776–780.	384 1965 CE	Radiographic study of birth cohorts. Anonymised CT data.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
6	Ali S, et al. The prevalence of spina bifida occulta in a Pakistani population: a study of dry human sacra. Anaesth, Pain Intensive Care. 2014;18: 157–161.	200 1954 CE	Modern anatomical study of archived dry human sacra.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
7	Shin SH et al. Spina bifida occulta: not to be overlooked in children with nocturnal enuresis. Int J Urol. 2013;20: 831–835.	160 1999 CE	Radiographic study in live patients with associated pathology.	Well-structured numerical results. SSBO and enuresis data separated Clear, accurate and included segment data for SSBO, S1

(n=)	Publication	Sample Size + Date	Characteristics	Risk of Bias Assessment*
8	Wu L et al. Variable morphology of the sacrum in a Chinese population. Clin Anat. 2009;22: 619–626.	203 1961 CE	Modern anatomical study of archived dry human sacra.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
9	Cakiroglu B et al. The adverse influence of spina bifida occulta on the medical treatment outcome of primary monosymptomatic nocturnal enuresis. Archive Italian Urol. 2014;86: 270–273.	233 1999 CE	Radiographic study in live patients with associated pathology.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
10	Solomon LB et al. Secular trend in the opening of the sacral canal: An Australian study. Spine. 2009;34: 244–248.	200 1945 CE	Radiographic study of birth cohorts. Anonymised CT data.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
11	El-Din A et al. Congenital anomalies of the vertebral column: a case study on ancient and modern Egypt. Int J Osteoarchaeol. 2006;16: 200–207.	270 2424 BCE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
12	Maat GJ et al. Analysis of human skeletons from a Hellenistic period, buried at a ruined Bronze Age building on Failaka, Kuwait. Maison de l'Oreint. 1990;18: 85–102.	12 1770 BCE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
13	Kim DW et al. Morphological diversities of sacral canal in children: three-dimensional computed tomography study. Korean J Pain. 2014;27: 253–259.	143 1996 CE	Radiographic study in live patients with associated pathology.	Well-structured numerical results. SSBO data and other anomaly data separated Clear, accurate and included segment data for SSBO, S1
14	Wu JW et al. Prevalence of spina bifida occulta and its relationship with overactive bladder in middle-aged and elderly Chinese people. Int Neurouro J. 2016;20: 151–158.	1061 1954 CE	Radiographic study in live patients with associated pathology.	Well-structured numerical results. SSBO and bladder dysfunction data separated Clear, accurate and included segment data for SSBO, S1
15	Fidas A et al. Prevalence and patterns of spina bifida occulta in 2707 normal adults. Clin Rad. 1987;38: 537–542.	2707 1911 CE	Radiographic study in live patients with associated pathology.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1

Table 8 (cont.)

(n=)	Publication	Sample Size + Date	Characteristics	Risk of Bias Assessment*
16	Shore LR. Abnormalities of the vertebral column in a series of skeletons of Bantu natives of South Africa. J Anat. 1930;64: 206–238.	155 1945 CE	Archaeological study of recovered human remains. (Dry human sacra)	Anecdotal data Clear, accurate and included segment data for SSBO, S1
17	Masnicova S et al. Developmental anomalies in skeletal remains from the great Moravia and Middle Ages cemeteries at Devin, (Slovakia). Intl J Osteoarchaeol. 2003;13: 266–274.	150 1115 BCE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
18	Hussien FH et al. Spinal pathological findings in ancient Egyptians of the Greco-Roman period living in Bahriyah Oasis. Int J Osteoarchaeol. 2009;19: 613–627.	77 289 BCE	Archaeological study of recovered human remains. (Dry human sacra)	Anecdotal data Clear, accurate and included segment data for SSBO, S1
19	Mays S. Spondylolysis, spondylolisthesis, and lumbo-sacral morphology in a medieval English skeletal population. Am J Phys Anthropol. 2006;131: 352–362.	422 1465 CE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results. SSBO and Spondylolysis data separate Clear, accurate and included segment data for SSBO, S1
20	Kim Y et al. Lumbosacral defects in a 16th – 18th century Joseon Dynasty skeletal series from Korea. Biomed Res Int. 2018;28: 1–8.	198 1666 CE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
21	Schweitzer ME et al. Spina bifida occulta: incidence in parents of offspring with spina bifida cystica. Spine. 1992;18: 785–786.	177 1932 CE	Radiographic study in live patients with associated pathology.	Anecdotal data Clear, accurate and included segment data for SSBO, S1
22	McGrath M et al. Anatomical observations related to radiological findings in spina bifida -occulta of the lumbo-sacral spine. J Osteopath Med. 2004;7: 70–78.	40 1994 CE	Radiographic study specifically designed for SSBO.	Anecdotal data Clear, accurate and included segment data for SSBO, S1
23	Papp T et al. Changes of the lumbar spinal canal proximal to spina bifida occulta. An archaeologic study of clinical significance. Spine. 1994;19: 1508–1511.	104 367 CE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1

(n=)	Publication	Sample Size + Date	Characteristics	Risk of Bias Assessment*
24	Jankauskas R. Variations and anomalies of the vertebra column in Lithuanian palaeoosteological samples. Anthropol. 2001;39: 33–38.	633 1467 CE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
25	Merbs CF. Sagittal clefting of the body and other vertebral development errors in Canadian Inuit skeletons. Am J Phys Anthropol. 2004;123: 236–249.	218 1867 CE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results. SSBO and other anomaly data separated Clear, accurate and included segment data for SSBO, S1
26	Stewart TD. The vertebral column of the Eskimo. Am J of Anthropol. 1932;17: 123–136.	217 1990 CE	Archaeological study of recovered human remains. (Dry human sacra)	Anecdotal data Clear, accurate and included segment data for SSBO, S1
27	Eubanks J et al. Prevalence of sacral spina bifida occulta and its relationship to age, sex, race, and the sacral table angle. Spine. 2009;34: 1539–1543.	2866 1885 CE	Radiographic study specifically designed for SSBO.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
28	Sutow WW et al. Incidence of spina bifida occulta in relation to age. Am J Dis Child. 1955;90: 211–217.	540 1921 CE	Radiographic study specifically designed for SSBO.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
29	Albrecht TL et al. Radiographical method to access the prevalence of sacral spina bifida occulta. Clin Anat. 2007;20: 170–174.	53 1937 CE	Radiographic study specifically designed for SSBO.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
30	Karlin IW. Incidence of spina bifida occulta in children with and without enuresis. Am J Dis Child. 1935;3: 374–393.	75 1840 CE	Radiographic study in live patients with associated pathology.	Anecdotal data Clear, accurate and included segment data for SSBO, S1
31	Jozsa L et al. The occurrence of spina bifida occulta in medieval and contemporaneous Hungarian populations. Anthropol Hunarica. 1992;22: 51–60.	233 1328 CE	Archaeological study of recovered human remains. (Dry human sacra) + Radiographic study specifically for SSBO.	Anecdotal data Clear, accurate and included segment data for SSBO, S1
32	Avrahami E et al. Spina bifida occulta of S1 is not an innocent finding. Spine. 1994;19: 12–15.	1200 1949 CE	Radiographic study specifically designed for SSBO.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1

Table 8 (cont.)

(n=)	Publication	Sample Size + Date	Characteristics	Risk of Bias Assessment*
33	Piontek J. Variation in the level of closure in the sacral canal of man. Folia Microbiol. 1971;4: 459–464.	316 1911 CE	Modern anatomical study of archived dry human sacra.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
34	Kubauat DM et al. A study of non-fusion of laminae of the first sacral vertebrae in Western India. Int J Recent Trends Sci Tech. 2013;6: 122–124.	302 1953 CE	Archaeological study of recovered human remains. (Dry human sacra)	Anecdotal data Clear, accurate and included segment data for SSBO, S1
35	Groza VM et al. Frequency of spina bifida occulta and other occult spinal dysraphism's in the medieval population of Isas city: skeleton palaeopathology in the necropolis discovered in the eastern part of the Princely Court, 17th century. Biol Anim. 2012;58: 195–204.	28 1660 CE	Archaeological study of recovered human remains. (Dry human sacra)	Anecdotal data Clear, accurate and included segment data for SSBO, S1
36	Henneberg RJ et al. Variation in the closure of the sacral canal in the skeletal sample from Pompeii, Italy, 79AD. Perspect Hum Bio. 1999;4: 177–188.	124 79 CE	Archaeological study of recovered human remains. (Dry human sacra)	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
37	Singh R. Classification causes and clinical implications of sacral spina bifida occulta in Indians. Basic Sci Med. 2013;2: 14–20.	140 1953 CE	Archaeological study of recovered human remains. (Dry human sacra)	Anecdotal data Clear, accurate and included segment data for SSBO, S1
38	Al-Dahhan MH et al. Evaluation of spina bifida occulta in young patients presented with lower back pain. Eur J Mol Clin Med. 2020;10: 4416–4422.	180 2016 CE	Radiographic study in live patients with associated pathology.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1
39	Kumar P et al. Spina bifida occulta in functional enuresis. Indian J Paediatr. 2005;72: 223–225.	48 1997 CE	Radiographic study in live patients with associated pathology.	Well-structured numerical results Clear, accurate and included segment data for SSBO, S1

Table 9. Raw data from frequency analysis

Publication	Date	Total SS	Male SS	Female SS	S1-S5	S1	S1-S2	S1-S3
El-Din and El Banna 2006	-2424	270			0.74			
			135		0.74			
				135	0.74			
2. Maat et al. 1990	-1770	12			8.30			
3. Molto et al. 2019	-874	116			5.17	13.79	0.86	
			64		7.80	10.93	1.56	
				52	1.92	17.30		
	-860	77				2.59		
	-868	193			3.10	9.32	0.51	
			23			8.69		
	1766	144				11.11		
4. Hussein et al. 2009	-289	77			54.54			
			41		51.21			
	124	119		35	60.00			
	124	119			4.2	16.80	3.36	
			56		7.4	19.64	3.57	
	110	130		63	1.58	14.28	3.17	
	110	130			0.76	6.15	3.84	0.76
			47		2.12	6.37	8.51	
				83		6.02	1.20	1.20

Percentage values for each sacral segment recorded. Publications 1–4. SS=Sample Size.

Table 9 (cont.)

Publication	Date	Total SS	Male SS	Female SS	S1-S5	S1	S1-S2andS4-S5	S1andS3-S5
5. Mays 2006	1465	422			1.18	4.50	0.23	
	1515	115				82.60		
6. Zemirline et al. 2013	768	30			3.33	6.66		
7. Henneberg and Henneberg 1999	39	124				13.46		
8. Papp and Porter 1994	367	104				13.46		
	617	27				29.62		
	967	77				10.38		
9. Masnicova and Benus 2003	1115	150					2.00	
			61				3.27	
				65			1.47	
	865	76					3.94	
			38				5.26	
				28			3.57	
10. Jankauskas 2001	1467	633			2.21	12.95		
11. Jozsa et al. 1992	1328	233			1.71	0.42		
12. Groza et al. 2012	1660	28					3.47	
	1662	129						0.77
			62					1.61
				62				1.09
	1670	91			1.09			1.09
			66		1.51			
13. Kim et al. 2018	1666	198			0.50	4.04		
			81		1.23	7.40		
				68		1.47		

Percentage values for each sacral segment recorded in each publication. Publications 5–13.

Table 9 (cont.)

Publication	Date	Total SS	Male SS	Female SS	S1-S5	S1	S1-S2	S1andS4-S5
14. Saluja 1988	1816	112			1.78	11.60	1.78	
	1928	140			2.14	10.00	2.85	
15. Merbs 2004	1867	218				43.11		
16. Solomon et al. 2009	1945	200				10.50	8.00	
			100			17.00	13.00	
				100		4.00	3.00	
	1985	200				16.50	9.50	
			100			23.00	15.00	
				100		10.00	4.00	
17. Stewart 1932	1900	217			2.76			8.29
			107		4.67			8.41
				96	1.04			9.37
18. Avrahami et al. 1994	1968	273				24.24		
			137			28.46		
				136		20.58		
	1958	259				22.77		
			131			27.48		
				128		17.96		
	1948	248				19.35		
			128			25.00		
				120		13.33		
	1938	229				8.73		
			111			8.10		
				118		9.32		

Percentage values for each sacral segment recorded in each publication. Publications 14–18.

Table 9 (cont.)

Publication	Date	Total SS	Male SS	Female SS	S1-S5	S1	S1-S2	L5-S5
Avrahami et al. 1994 (cont)	1928	191				6.80		
			93			7.52		
				98		6.12		
19. Eubanks and Cheruvu 2009	1885	2866			1.22		11.13	
20. Lee et al. 2011	1965	384				2.34		
			194			2.57		
				190		2.10		
21. Karlin 1935	1913	75			12.00	62.00	16.00	12.00
22. Shore et al. 1932	1840	78				3.84		
23. Sutow and Pryde 1955	1945	155				51.61		9.03
			86			46.51		10.46
				69		57.97		7.24
	1941	95				44.21		6.31
			44			47.72		9.09
				51		41.17		3.92
	1936	108				39.81		4.62
			48			52.08		10.41
				60		30.00		
	1921	182				14.83		2.19
			79			54.43		3.79
				103		20.38		0.97
	1902	46				23.91		
	1902	87				16.57		
	1902	87				22.18		

Percentage values for each sacral segment recorded in each publication. Publications 19–23.

Table 9 (cont.)

Publication	Date	Total SS	Male SS	Female SS	S1-S5	S1	S1-S2	L5-S2
24. Singh 2013	1953	140			3.57			
25. Kubauat et al. 2013	1953	302				10.92		
26. Schweitzer 1992	1932	177						15.81
			32					15.62
				37				13.50
			53					16.98
				56				16.07
27. Cakiroglu et al. 2014	1999	233						1.28
			151			22.31	0.85	
				72		3.31	0.66	4.16
						65.27	5.55	
28. Mith and Tayles 2004	1994	40				22.50		
			20			22.22		
				20		27.77		
29. Piontek 1971	1911	316			1.26	3.48	3.48	1.26
			187		1.60	2.67	4.81	1.60
				129	0.77	4.65	1.55	0.77
30. 0 et al. 2004	1954	200			4.50		3.50	
31. Kumar et al. 2005	1997	48				16.66	4.16	10.41
32. Kim 2014	1996	143					15.40	
33. Wu et al. 2009	1961	203			2.95		18.20	
34. J.W. Wu et al. 2016	1954	1061				11.96	0.65	

Percentage values for each sacral segment recorded in each publication. Publications 24–34.

Table 9 (cont.)

Publication	Date	Total SS	Male SS	Female SS	S1	L5-S5
35. Fidas et al. 1987	1941	570			28.59	
			301		37.20	
				269	18.95	
	1931	877			20.98	
			411		29.68	
				460	13.47	
	1921	658			20.97	
			333		23.72	
				325	18.15	
	1911	380			20.58	
			208		25.00	
				172	15.11	
	1901	173			16.76	
			80		16.25	
				93	17.20	
	1891	52			19.23	
			24		20.83	
				28	17.85	
36. Shin et al. 2013	1999	160			16.25	
37. Urrutia et al. 2016	2005	228			35.08	
38. Al-Dahhan et al. 2020	2016	180			18.5	4.40
39. Albrecht et al. 2017	1937	53			1.25	

Percentage values for each sacral segment recorded in each publication. Publications 25–39.

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