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Prevalence of wormian bones worldwide: a critical review

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Abstract: Wormian bones (WB) are the irregular bone structures developed from additional centers of ossification. Although they are commonly found in healthy individuals, under certain conditions (number >10, mosaic pattern, large size), they can indicate pathology. While their coexistence with numerous diseases is well-documented, and various studies have reported their prevalence in populations of various geographic regions, no qualitative critical review of such studies has been conducted. The aim of this paper is to perform a critical review of research studies on the presence of Wormian bones in populations worldwide, with a particular emphasis on the methodology used and the selection of the samples studied.

A sample of 44 original research articles was selected via PubMed and Google Scholar databases. Four criteria were assessed: 1) number of individuals in each group, 2) known sex of individuals, 3) selection criteria of individuals, and 4) implementation of the statistical analysis. The origin of the research sample was determined as well as the method of the WB calculation, and data on the WB prevalence worldwide was collected in tabular form.

The reported size of the research samples varies from 22 to 628 individuals, derived from both contemporary and archaeological populations. Four major formulas were used in order to provide the frequency of WB. The sex of individuals was known in 18 (40.9%) articles. Most of the articles focused on Asian samples.

The difficulties in comparing data on the Wormian bones are caused by considerable inconsistency in the methodology used to research this phenomenon. Therefore, the interpopulation comparisons currently made may not be correctly estimated. Our study highlights the need for using more comprehensive and consistent data collection as well as processing protocol suitable for populational research on sutural bones.

Key words: cranium, skull, suture, sutural bone, Wormian bone.

Original article

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Introduction

The human skull consists of two developmentally and functionally distinct groups of bones: viscerocranium and neurocranium (Reicher and Łasiński 2010). The number of skull bones is generally constant, but in some cases, the number might increase due to the presence of so-called Wormian bones (WB) which are also known as 'intrasutural bones', 'sutural bones', 'skull accessory ossicles', 'supernumerary ossicles', 'intercalated bones', 'accidental bones', 'intercalary ossicles', '*Schaltknochen*', '*ossa Wormiana*', '*ossa suturalia*' (Goyal et al. 2019; Nowak et al. 2018; Rajni et al. 2018; Murlimanju et al. 2011). In particular cases – when the bones occur in the position fontanelles (Natsis et al. 2019; Nikolova et al. 2016) – they have specific names such as 'bregmatic bone' and 'Kerckring bone' (in bregma, Nikolova et al. 2016; Vishali et al. 2012), or 'pterion ossicles' and 'epipteric bone' (in pterion, Standring 2016). There is also a large bone on the posterior part of the skull (upper area of the occipital squama) referred to as the 'Inca bone' (Cirpan et al. 2014) or 'interparietal bone'.

Wormian bones are irregular, diversiform bone structures developed from the additional centers of ossification (Nowak et al. 2018; Bellary et al. 2013) and interjected in the cranial sutures. They are present not only in modern humans but also in the archeological context (Nikolova et al. 2014; Panzer et al. 2014), in Neanderthals (Bruner 2004), and animals (Zambrano et al. 2021; Smith et al. 1977).

The size of a single accessory bone can vary from less than 1 mm x 1mm up to more than 5 cm x 9 cm in width and length (Sreekanth and Samala 2016). In addition, these bones have multiple shapes: oval, round, oblong, quadrilateral, and irregular (Parker 1905, as cited in Murlimanju et al. 2011). Wormian bones are generally a feature of the neurocranium and occur mostly in the lambdoid suture, especially on the right side (Nowak et al. 2018; Bellary et al. 2013). Other frequent localizations are coronal suture, bregma, lambda, pterion, and asterion. However, WB may, although rarely, occur in the sutures connecting the craniofacial bones – for example in the frontonasal suture (Edwards et al. 2017) or in the orbital cavity between the frontal, lacrimal and ethmoid bone (Rizvi et al. 2018). The anatomical denomination of Wormian bones is still debated (Romero-Reverón 2020) while the etiology of sutural bone formation still remains unclear. It has been hypothetised that WB might indicate genetic factors (Finkel 1975; Bennett 1965), mechanical pressure on the skull bones in early stages of ontogenetic development, such as artificial cranial deformations (Sanchez-Lara et al. 2007; O'Loughlin 2004; El-Najjar and Dawson 1977), metabolic disorders (Hess 1946, as cited in Jeanty et al. 2000), as well as environmental factors (Barberini et al. 2008; Sanchez-Lara et al. 2007). Currently, more holistic interpretations are adopted that recognize the influence of all the aforementioned factors in WB development (Di Ieva et al. 2013; Barberini et al. 2008). Some studies suggest that the presence of sutural bones might indicate developmental instability (Di Ieva et al. 2013; Vishali et al. 2012; Barberini et al. 2008).

The Wormian bones have been reported to commonly occur in healthy populations, and their presence typically is not associated with any pathological conditions (Natsis et al. 2019; Andrade et al. 2018; Johal et al. 2017; Walulkar et al. 2012); however, their significant number (above 10), size (more than 6 mm x 4 mm), or characteristic mosaic pattern are clinically considered as indicators of several congenital diseases, mostly osteogenesis imperfecta (Cremin et al. 1982) exhibited by abnormally numerous Wormian bones (Semler et al. 2010). Other diseases are also frequently associated with WB, such as hypophosphatasia, craniosynostosis, hypothyroidism, cleidocranial dysostosis, rickets, pyknodysostosis (osteopetrosis acro-osteolytica), pachydermoperiostosis, congenital hypothyroidism, hydrocephalus, otopalatodigital syndrome, Hajdu-Cheney syndrome, Menkes syndrome (Ratnaningrum 2020; Saylisoy 2020; Basnet et al. 2019; Romero-Reverón and Arráez-Aybar 2019; Kumar and Ratna Prabha 2016; Marti et al. 2013). Many of them are congenital disorders or CNS (central nervous system) anomalies. Studies have shown that Wormian bones can help in diagnosis and treatment of those conditions (Goyal et al. 2019; Romero-Reverón and Arráez-Aybar 2019; Jeanty et al. 2000). As Wormian bones are developed prenatally (Jeanty et al. 2000), they can be detected during the routine USG examination, and used as the prenatal diagnosis of severe or lethal conditions (Tonni et al. 2013).

Wormian bones can be also considered as important from the medicolegal perspective as their characteristic pattern, can be used for identification of an individual (Kuharić et al. 2011; Jayprakash 1997); moreover, due to their association with abnormal bone brittleness, WB can prove critical in the diagnosis of the children's bone trauma in order to exclude the possibility of physical abuse (Johal et al. 2017; Govsa et al. 2014; Marti et al. 2013). During X-Ray examination of the head, Wormian bones can also be mistaken for fractures (Narayan et al. 2019; Romero-Reverón 2017). Radiologists and surgeons have been advised to take WB into account while planning surgical interventions to the head to avoid possible injury (Ratnaningrum 2020; Kumar and Ratna Prabha 2016).

Interest in Wormian bones in the last decade has been significantly growing (see fig. 1).

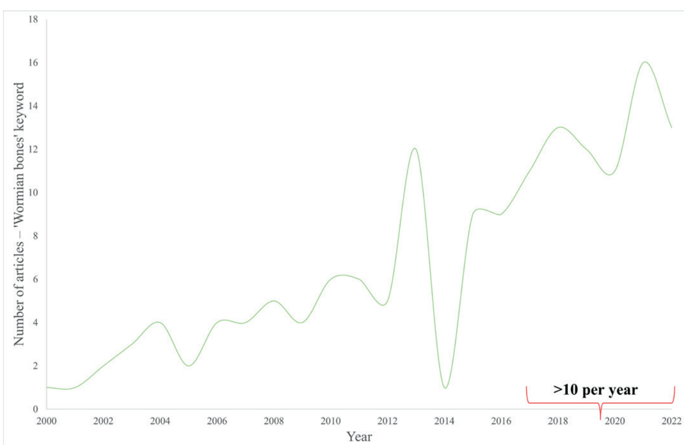

Fig. 1. Number of articles published between 2000 and 2022, found in PubMed by the 'Wormian bones' keyword (full texts only)

Some studies have discussed the association of WB with various diseases (case reports), while other have established the relationship between WB and biological phenomena, such as metopism (Li et al. 2022; Cirpan et al. 2016), individual genes or groups of genes that influence WB formation (Zimmerman et al. 2019; Kague et al. 2016), and variants of the skull morphology (Basnet et al. 2019). Since WB are considered as an anthropological marker of the interpopulation distance (Natsis et al. 2018; Gümüsburun et al. 1997; Pal and Routal 1986), some studies report the WB prevalence in human populations worldwide. In many cases, the data presented and juxtaposed therein are further cited by researchers; however, so far, no qualitative critical analysis of this group of articles has been carried out. Such a comprehensive review of the above studies, however, would be very useful for other researchers by providing an established basis for future research on Wormian bones.

The aim of this study is to review articles on the presence of Wormian bones in populations worldwide, with a particular emphasis on the methodology used and the selection of the study sample. In this study, the term 'Wormian bones' is used predominantly due to its common occurrence in the reviewed articles.

Material and Methods

This study is a semiqualitative review of literature. By using 'Wormian bones' as the keyword search term, 208 articles in the PubMed database were found. After adding the filter for 'full text', the total number was 169 (August 22, 2022).

In addition, in order to supplement the study material, the authors also used the Google Scholar database. Overall, the number of records found using the same 'Wormian bones' search term was 5870 (citations and patents were not taken into account). To limit the scope of the material, the following search criteria were used:

1. Published in the last 10 years (2012–2022)
2. 'Highest relevance' of the search filter selected

The total sum of records found using the above search criteria was 2600 (August 22, 2022). Since the search with the 'highest relevance' option was used, the further review was limited to the first five pages of Google Scholar results (50 articles), considering them to be the most relevant to the search criteria.

The total number of records obtained at this stage of search (preliminary database) was 219.

At the following stage, the articles were reviewed in terms of meeting the following selection criteria:

1. Research on Wormian bones (or their specific categories such as pteric bones, etc.) in any context (assessed by using the 'Wormian bone' or 'sutural bone' term in the title, keywords or abstract) conducted on human sample
2. Article available online
3. Full text of article available free (Open Access) or via institutional approach of authors' units
4. Article in English, Polish, German or Spanish.

After excluding duplicates (publications recorded both in PubMed and Google Scholar databases) the total number of articles was 176.

Such obtained material was then divided into six groups/categories: 1) case reports, 2) original articles on clinical approaches, 3) medicolegal approaches,

4) original articles on Wormian bones prevalence in populations worldwide, 5) reviews, 6) varia (i.e., articles directly or indirectly related to Wormian bones, which were not assigned to any of the above category). Only the fourth group (original articles on Wormian bones prevalence in populations worldwide) was selected as the subject of this research. An article was assigned to this group if it indicated the studied population in the title, introduction, or in the description of the research material. Articles that investigated the relationship between WB and skull deformities in various populations were excluded from the study. The preliminary number of articles included into the fourth group was 39.

In order to enrich the material, the authors checked the reference lists of these articles and selected additional publications regarding the WB prevalence in particular populations that were not included during the previous selection steps. Although such identified articles were only few, they report relevant results that are cited in further research as reference data. Because this study is, to our knowledge, the first attempt to propose a semiqualitative review of the WB issue, the chosen articles were not selected for peer review, citation score, impact factor, or other parameters. The final number of articles in the fourth group was 44 (see fig. 2), published between 1975 and 2022.

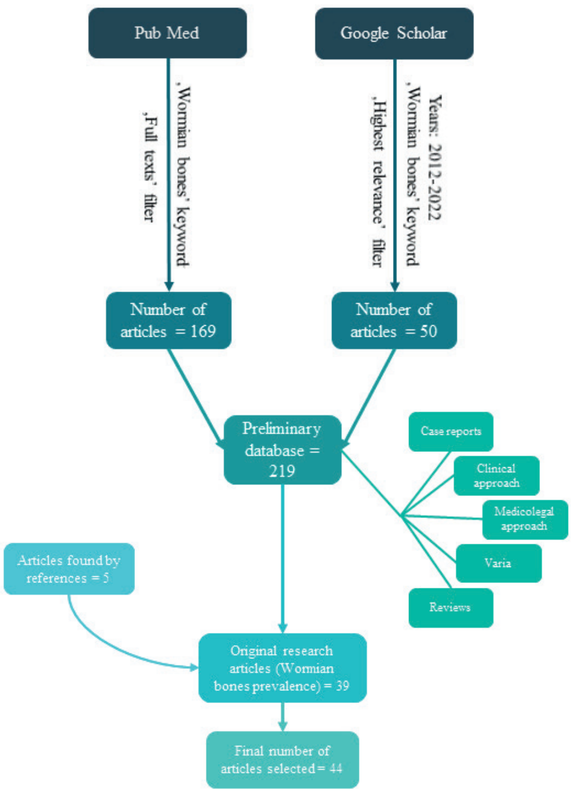

Fig. 2. Articles selection process

A scoring system, inspired in part by the Bradford-Hill criteria (Rothman and Greenland 2005), was proposed for the assessment of the evidential strength of the analyzed articles. Four benchmarks were selected, and one point was noted for each criterion:

1. ≥30 individuals in each examined group
2. Known sex of individuals
3. Statistical testing (regardless of the type of test selected)
4. Discussed selection criteria of the material

The data was tabulated according to the point category: 0–4 points. Central tendency measures were calculated using the STATISTICA 13.3 software (TIBCO; no. of license: JPZ007B482801A-RACD-9).

Results

Due to the scoring system implemented and discussed above, the division of the articles from the group of articles selected for review is as follows: 0 points – 1 article; 1 point – 11 articles; 2 points – 15 articles; 3 points – 12 articles; 4 points – 5 articles (table 1). The reported size of research samples varies from 22 to 628 individuals (*mean = 125, Me = 79, Mo = 50, Q1 = 50, Q3 = 155.5*). Research samples used in these studies were collected from contemporary institutions (e.g., post-sectional dry skulls, exhibits of medical museums with the number of articles of 36), or historical populations (e.g., from archaeological excavations or museums with a number of 5 articles), or mixed sources with only one article. In a few cases the origin of samples was unclear. Where possible, the authors determined the origin of the sample on the basis of the condition of the skulls visible in the photographs. In 18 studies (40.9%), the sex of the subjects was known. The age of individuals was known in 33 (75%) of all examined articles, although in most cases they were described simply as 'adults'. In two articles, the division of individuals into age categories was given as follows:

1. Natsis et al. 2019 – three age groups: 20–39 (n = 41), 40–59 (n=30), over 60 (n = 95).

2. Durge 2016 – four age groups: 20–30 (male n = 18, female n = 21, total n = 39), 30-45 (male n = 32, female n = 26, total n = 58), 45-60 (male n = 10, female n = 21, total n = 31), over 60 (male n = 12, female n = 20, total n = 32).

Some articles were thematically limited to selected points of the skull: asterion – 3 articles; – pterion – 7 articles; asterion and pterion – 1 article; preinterparietal/interparietal/Inca bones – 4 articles. In two cases, the preinterparietal/interparietal/Inca bones were excluded from the study.

The described research was conducted mainly in the area of South Asia (see fig. 3) with a high representation of data from India.

In 24 out of 44 articles (54.5%), a value of general prevalence of Wormian bones in a particular population was reported. This value varied from 4.7% in Rajasthan (Masih et al. 2013) to 88.8% in South India (Nayak and Shetty 2019). Moreover, the articles provide the WB frequency in particular skull sutures (table 2) revealing that the site of the most frequent occurrence of these structures was the lambdoid suture, which corresponds to other studies (El-Najjar and Dawson 1977; Romero Reverón 2017). The next most common locations for sutural bones were the asterion (79.7%

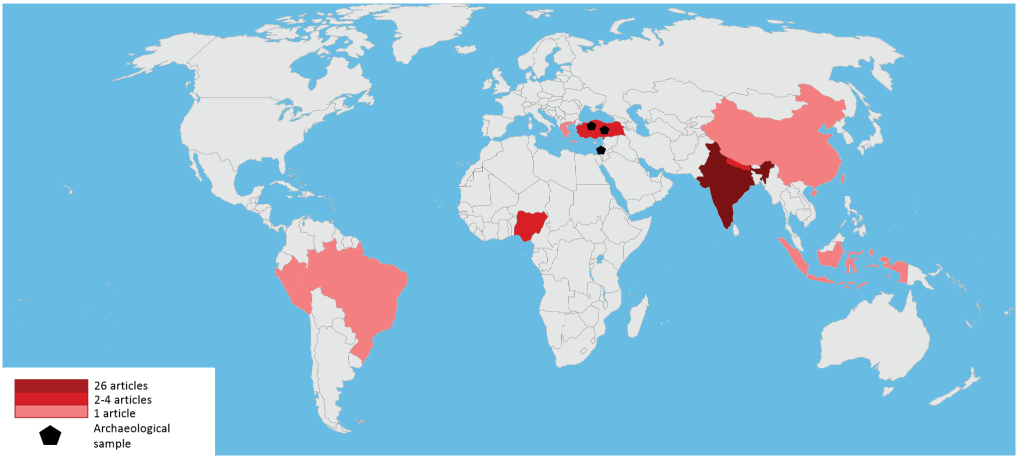

Fig. 3. Geographical distribution of research on Wormian bones (blank map by Ian Maphy, the authors' modification)

in South Indian population, Lekshmy et al. 2017), coronal suture (66.6% in South Indian population, Nayak and Shetty 2019), and the squamous suture (45.5%, also in South Indian population, Lekshmy et al. 2017) – the highest percentages recorded are given in this study. The prevalence of preinterparietal or interparietal or Inca bones occurrence varies from 1.3% in Central India (Marathe et al. 2010) to 13.3% in South India (Nagarajan and Ganesh 2017). All percentages listed indicate the number of individuals with a Wormian bone in specific cranial site (suture), relative to the entire study group.

26 articles (59.1% of reviewed sample) relate to the Indian population. Articles investigating populations from other areas of Asia taken into account included Nepal = 2, Indonesia = 1, Turkey (including Anatolia) = 6, China = 1, and Israel (Lachish) = 1. Data on Asian populations (including archaeological) account for over 84% of the sample. The remaining articles concern Greek (1 article), Nigerian (4 articles), Brazilian (1 article), Peruvian (1 article).

Discussion

The assessment of the variability in the occurrence of biological phenomena in a population is the first and indispensable step towards analyzing the etiology of any given phenomenon. The collected and available data on the variability of the phenomenon in other populations allow for comparative analyzes. Researchers investigating Wormian bone in the analyzed populations often refer to tabulated data from other studies (Li et al. 2022; Sah et al. 2017; Kalthur et al. 2017; Ghosh et al. 2017; Gümüsburun et al. 1997). In the above studies, the researchers refer to the global ranges of variability determined by the populations with the lowest and highest percentage of sutural bones – such as: "The reported incidence is variable, ranging from around 10% (in Caucasian skulls), through 40% (in Indian skulls), to 80% (in Chinese skulls)" (Khan et al. 2011).

This study reviews the original articles that reported the incidence of Wormian bones in populations from different geographic regions in order to create an organized database; however, the difficulty in completing such a database results from the inconsistency of the methods used to assess the presence of the Wormian worldwide. The most considerable differences in the adopted methodology concern calculating the frequency (percentage) of a feature in the analyzed publications. Four dominant strategies were shown (table 2):

1. WB frequency as the quotient of the sum of all Wormian bones (in specific sites of the skull) counted in the study sample and the entire study group (WB-positive and WB-negative individuals, divided according to sex, if stated), multiplied by 100% This strategy has been used in 3 (6.8%) cases.
2. WB frequency as the quotient of the sum of all Wormian bones (in specific sites of the skull) counted in the sample and the number of WB-positive individuals (divided into sex groups, if distinguished), multiplied by 100%. This strategy has been used in 2 (4.6%) cases.
3. WB frequency as the quotient of the number of individuals with WB (in specific sites of the skull) and the entire study group (WB-positive and WB-negative individuals, divided into sex groups, if distinguished), multiplied by 100%. This strategy has been used predominantly, in 31 (70.5%) cases.
4. WB frequency as the quotient of the number of individuals with WB (in specific sites of the skull) and the group of WB-positive individuals (divided into sex groups, if distinguished), multiplied by 100%. This strategy has been used n 5 (11.4%) cases.

In 5 cases, either a different way of calculating attendance was used or it was not specified how the result was obtained. The inconsistency of the calculation methods in which the result is reduced to one value category – the percentage (%) – creates a risk that the data obtained and compiled with each other may be under- or overestimated. Inaccuracies also apply to the classification of the Wormian bones themselves; for example, (pre-)interparietal/Inca bones are treated as a separate category or included into the group of lambda bones or sagittal/lambdoid sutures.

Of the selected studies, 2 were conducted on archaeological samples: Lachish (Finkel 1975) and Anatolian (Gümüsburun et al. 1997). These two studies are relatively often quoted in tabular summaries, in which they are compared with contemporary samples (Li et al. 2022). However, this approach may raise concerns because genetic distances between historical populations may differ considerably compared to those derived from contemporary populations. Moreover, a direct comparison of modern trials with groups that are significantly distant in time, such as the Lachish population analyzed in the article by Finkel (1975) dated as the late bronze age, may omit some aspects of morphological changes related to the process of human microevolution. For this reason, a greater precision in the description of the tested sample is important. One of the included in this review articles focused on the occurrence of morphological variants of pterion in mixed archaeological (13-century Byzantine) and contemporary (20-century Turkish) sample (Illknur et al. 2009). This study states that the pteric bone occurs in 6.25% od Byzantine sample and in 3.6% of the modern population. However, the statistical significance of differences in these both frequencies was not tested, and the samples were relatively small (28 modern skulls and 16 Byzantine skulls), so the obtained results may be unreliable (Illknur et al. 2009).

In 16 studies included in this review, the origin of the sample was not specified. It is also worth noting is that in some studies osteological materials from anatomical museums at medical faculties were used. The origin of such materials should also be carefully explained because museums can store both prepared contemporary remains and historical bone material (i.e., those from archaeological excavations including modern, medieval, and even older remains) in their collections.

Modern samples may not only differ from archaeological samples in terms of genetic distance, but also be expoused to different environmental conditions, exhibit different health status which may, potentially, affect the incidence of Wormian bones and the high number of which is associated with numerous diseases. Furthermore, historical material from archaeological excavations or crypts is subject to taphonomic changes. For example, an excavated material may be damaged or dehydrated in such a way as to posthumously unseal the space of the cranial sutures. This poses a risk of accidentally losing Wormian bone post mortem. Such a risk is much smaller in comparison with contemporary post-sectional samples. In historical populations, the structure of sex and age is also different, which may distort the interpretation of data.

In 26 out of 44 (59.1%) analyzed articles, the researchers did not specify the sex of the examined remains. The omission of such information makes it difficult to undertake further works on the study of sexual dimorphism, on which there is no consensus in the literature (Natsis et al. 2019; Cirpan et al. 2016; Patil and Sheelavant 2012). Some authors argue that there are no differences in the prevalence of WB between sex groups. However, according to the 16 discussed articles in which the sex of individual was determined (research on male group only excluded), it may appear that the Wormian bones are more common in males (table 3). Note, however, that these results may represent non-statistical observations, not based on a significant difference testing result.

In the four included in this review articles (4 of the 44, 9.1%), the examined sample consisted less than 30 individuals, which are considered minimum that allows inference on quantitative data. None of these studies attempted to estimate the required sample size. The collection of research material depends on many substantive factors, such as the total amount of osteological material obtained through archaeological excavations, non-substantive factors such as funds, consent to share the material, and the duration of the study. In population studies, providing a sample size that allows for statistical analyzes, although critical, was implemented only in 23 out of the 44 (52.3%) evaluated articles. Statistical inference with respect to compiling numerical data from population studies is the only way to distinguish significant biological relationships from numerical randomness. Without which it is rather difficult, if not impossible, to properly estimate the differences in the occurrence of Wormian bones between sex groups.

Of the 44 original articles included in the analysis, more than a half (26 of the

44, 59.1%) relate to the Indian population. Nevertheless, there is no consensus on the reported frequency of suture bones in populations from the same areas; consider the following examples: in the East Indies reported from 29.7% (Yadav and Salam 2020) to 72.3% (Purohit and Yadav 2019), in North India – from 35.3% (Goyal et al. 2019) to 51% (Rajni et al. 2018). These discrepancies may result from the large diversity of the populations inhabiting the area of India but also from the methodology adopted in the assessment of the Wormian bones.

Studies regarding populations from Asia account for over 84% of the sample collected according to the proposed method partly based on indications and systematic reviews of publications in established scientific databases. In contrast, there is an insufficient number of studies on populations from Europe, Africa, Australia, Oceania as well as North and South America. The problem of older research on WB, published in print (not digitized) in languages other than those used internationally, also should be mentioned.The lack of digital and linguistic access for researchers who want to undertake reviews or comparative analyzes can create considerable gaps in the methodology and results. An example of such materials could be the publication of Česnys and Balčiūnienė (1988) on Lithuanian populations; therefore, it is important to enrich available databases of scientific articles with archival works on the subject. Translated non-English-language studies and English-language meta-analyzes published in native languages concerning the prevalence of Wormian bones, as well as other morphological and anatomical studies in local populations, should be made available to the worldwide scientific and scholarly community.

Due to the major inconsistency regarding research methods used to determine the frequency of Wormian bones, comparing archival data may not be reliable and therefore existing knowledge regarding this phenomenon may not reflect the actual biological regularities. Consequently, there is a need for clarity regarding the description of the data collection protocol which would allow conducting a more robust meta-analysis and further research on Wormian bones. In addition, there is a significant gap in the data on the presence of sutural bones in non-Asian populations which should encourage research on materials from these regions of the world.

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Conflict of interests

The authors declare no conflict of interests

Authors' contributions

AB is the head of the research team, originator, main researcher, author of the working and final version of the paper, performer of statistical analyzes and interpretation of calculation results. RR-R is the co-creator of the draft and final version of the work and the substantive supervisor.

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Table 1. Research Characteristics

Article	Population	Sample	No. of Individuals	Known Sex and Age	Research Method	Comments
			SCORING = 0			
Ahad and Thenmozhi, 2015	South Indian	cont?	25	A	Asterion bones counted and measured relative to cranial land-marks (bilaterally)	asterion only
			SCORING = 1			
Saxena et al. 1986	Nigerian	cont?	40	A	Preinterpalietal bones counted	preinterpatietal bones only
Singh 2012	Indian	–	55	S, A	Number of WB in asterion noted (bilaterally)	asterion only
Masih et al. 2013	North Indian (Rajasthan)	cont	150	S, A	Presence of WB noted	–
Durgesh et al. 2015	Indian (Andhra Pradesh)	cont?	48	–	Occurrence of WB noted	WB present only in the lambdoid suture
Praba and Venkatramani-ah 2015	Indian (Tamil Nadu)	cont?	50	S, A	WB counted in bregma, lambda, pterion and asterion (regardless of the side)	–
Raja and Siva 2016	South Indian	cont?	75	A	WB in pterion counted	pterion only
Veeresh et al. 2016	Indian	cont?	50	A	Number and location of WB not-ed (bilaterally); WB photographed	–
Sah et al. 2017	Nepalese	cont?	80	A	Presence and localization of WB noted	–
Uchewa et al. 2018	Nigerian	cont	22	S, A	WB counted and photographed; lateralization not included	–
Rajni et al. 2018	North Indian	cont/ cont (?)	55	A	Number and location of WB not-ed (bilaterally)	Material including cadaveric and muse-um samples
Ratnaningrum 2020	Indonesian	cont?	69	–	Presence, localization and varia-tions in WB shapes noted (bilat-erally)	–

SCORING = 2						
Illknur et al. 2009	Anatolia	cont/arch	44	S/–, A	Epipteric bones counted; measurements between pterion and several landmarks taken both in manual and digital way (on photographs)	pterion only; sex (male) assessed on archaeological sample only
Murlimanju et al. 2011	Indian	cont?	78	–	Incidence and topographical distribution of WB (bilaterally)	Interparietal and preinterparietal bones not considered as WB
Walulkar et al. 2012	Central Indian (Vidarbha)	cont	225	S, A	WB counted bilaterally, measured (max. width x max. length), incidence of various shapes of WB noted	–
Sudha et al. 2013	South Indian	cont	150	A	Epipteric and asterion bones noted bilaterally	pterion and asterion only
Eboh and Obaroefe 2014	Nigerian	–	50	–	Epipteric bones counted bilaterally; measurements between pterion and several cranial landmarks	pterion only
Ghosh et al. 2017	East Indian	cont	120	A	WB counted bilaterally; topographic distribution analysis, symmetry of WB occurrence analysis	–
Tallapaneni and Niveditha 2016	South-Eastern Indian (Telangana)	cont?	111	–	Presence of Wormian bones in respect to their location and number noted (bilaterally)	–
Uday and Ratna Prabha 2016	Indian (North Karnataka)	cont	200	–	Presence, localization and number of WB noted	–
Ukoha et al. 2013	Nigerian	cont	56	A	Frequency of pterion's types recorded according to Murphy (1956)	pterion only
Kalthur et al. 2017	South Indian (Karnataka)	cont?	50	S, A	Epipteric bones counted and measured relative to cranial landmarks (bilaterally)	pterion only

Article	Population	Sample	No. of Individuals	Known Sex and Age	Research Method	Comments
Lekshmy et al. 2017	South Indian	cont	200	A	WB counted (bilaterally); topographic distribution of WB	–
Nagarajan and Ganesh 2017	South Indian	cont	60	–	Presence and number of Inca bones; skulls with Inca bones photographed	Inca bones only
Narayan et al. 2019	Eastern Indian (Bihar)	cont?	30	A	Number of WB and their localization noted bilaterally; cranial index examined	–
Nayak and Shetty 2019	South Indian	–	27	S, A	WB noted (regardless of the side), photographs of skulls with WB	–
Yadav and Salam 2020	Eastern Indian	cont	64	S, A	WB counted (regardless of the side)	CT scans of head and neck
SCORING = 3						
Pal and Routal 1986	Indian (South Gujarat)	cont	117	–	WB noted bilaterally for each morphological type of skull; correlation with cephalic index examined	–
Gümüsburun et al. 1997	Anatolian-Ottoman	arch	302	S, A	WB counted bilaterally; examined correlation with the cephalic index (3 morphological forms)	–
Govsa et al. 2014	Turkish	cont	300	A	Presence, localization and number of WB noted bilaterally	3D reconstructions with volume rendering; preinterparietal bones treated separately
Oguz et al. 2004	Turkish	cont?	26	S (males only), A	Types of pterions (according to Murphy (1956)) noted; measurements between pterion and selected landmarks; bone thickness measurements	pterion only

Cirpan et al. 2014	West Anatolian	cont	151	A	Inca bones counted following the Hauser and De Stefano (1989) and Kadanoff and Mutafov (1968) classification	Inca bones only
Cirpan et al. 2016	West Anatolian	cont?	150	A	Presence, frequency and topographic distribution of WB assessed bilaterally; photography	–
Durge 2016	South-Easter Indian (Andhra Pradesh)	cont?	160	S, A	WB number and localization noted	–
Basnet et al. 2019	Nepalese	–	70	A	WB counted bilaterally; examined correlation with the cephalic index (3 morphological forms)	Inca bones treated separately
de Lucena et al. 2019	North-East Brazilian	cont	30	S, A	Numbers of WB in asterion noted; measurements between asterion and several skull landmarks	asterion only
Li et al. 2022	Chinese	cont	285	A	Width and length of WB measured, counted (bilaterally); types of WB shapes counted	Inca bones treated separately
Murrieta-Angulo et al. 2019	Peru	cont	90	S, A	Type of pterion bilaterally noted according to Murphy's classification (1956); measurements between the ossification center and zygomatic arch; photographs	pterion only
Shiv et al. 2020	Indian (Southern Haryana)	cont	130	S	Location of WB along the coronal, sagittal and lambdoid suture on ectocranial surface noted; photographs	–
SCORING = 4						
Finkel 1975	Lachish	arch	628	S	WB presence noted (in lambdoid, sagittal or coronal suture); skull metric differences in relation to WB presence/absence examined	Inca bones excluded

Article	Population	Sample	No. of Individuals	Known Sex and Age	Research Method	Comments
Goyal et al. 2019	North Indian (Haryana)	cont	147	–	Presence, number and topographic distribution of WB noted (regardless the side of the skull)	Interparietal and pre-interparietal bones not considered as WB
Marathe et al. 2010	Central Indian	cont	380	A	Incidence of Inca bone noted; measurements of Inca bone	Inca bones only
Natsis et al. 2019	Greek	cont	166	S, A	WB presence noted bilaterally and both exo- and intracranially; WB presence on viscerocranium noted; side asymmetry analysis	Inca bone excluded from the study
Purohit and Yadav 2019	East Indian	–	180	S, A	Presence and location of WB noted bilaterally; photographs of WB taken; cephalic index included	Inca bones not counted as WB

Abbreviations: arch – archaeological sample, cont – contemporary sample; S – sex, A - age. 'Known age' was determined positively both when the exact age categories were given or when subjects were assigned the general 'adult' age category. 'Bilaterally' description in the table refers to the parts of the skull (sutures or landmarks) that appear paired.

Table 2. Wormian Bones Prevalence Among Various Populations Worldwide

Article	Presence	Total no.	WB in main sutures					WB in specific sites of the skull						
			C	S	Sq-R	Sq-L	L	B	LL	A-R	A-L	P-R	P-L	I
SCORING = 0														
Ahad and Thenmozhi 2015 ⁂	–	–	–	–	–	–	–	–	–	12% / Σ: 32%	20%	–	–	–
SCORING = 1														
Saxena et al. 1986 ⁂	–	–	–	–	–	–	–	–	–	–	–	–	–	*II* single: 1 (2.5%) *PI* single: 4 (10%) multi: 1 (2.5%)
Singh 2012 ⁂	–	–	–	–	–	–	–	–	–	♂: 25.0 % ♀: 14.8%	♂: 10.7 % ♀: 14.8%	–	–	–
Masih et al. 2013 ⁂	♂: 6 (4.1%) ♀: 1 (3.6%) Σ: 7 (4.7%)	–	–	–	–	–	–	–	–	–	–	–	–	–
Durgesh et al. 2015 ⁂	24.1%	–	–	–	–	–	14	–	–	–	–	–	–	–
Praba and Venkatramaniah 2015	–	♂: 56 ♀: 27	–	–	–	–	–	♂: 0 ♀: 0	♂: 13 ♀: 1	♂: 35 ♀: 19		♂: 8 ♀: 7		–
Raja and Siva 2016 ⁂	–	–	–	–	–	–	–	–	–	–	–	1 / Σ: 3 (4%)	2	–

Article	Presence	Total no.	WB in main sutures						WB in specific sites of the skull					
			C	S	Sq-R	Sq-L	L	B	LL	A-R	A-L	P-R	P-L	I
Veeresh et al. 2016 ⁂	16 (32%)	–	–	0	–	–	R: 7 L: 15 Σ: 22 (44%)	0	3 (6%)	1 Σ: 3 (6%)	2	1 Σ: 1 (2%)	0	–
Sah et al. 2017 ⁑⁑	55 (68.7%)	–	0	2 (3.6%)	–	–	35 (63.6%)	0	–	11 (20%)		7 (12.7%)		–
Uchewa et al. 2018 ⁂	10 (45.5%)	–	0	0	–	–	8 (36.4%)	0	–	–	–	Σ: 2 (9.1%)		–
Rajni et al. 2018 *	28 (51%)	–	0	1 (2%)	2 Σ: 3 (5%)	1	6 Σ: 14 (26%)	8	4 (7%)	1 Σ: 3 (5%)	2	1 Σ: 1 (2%)	0	–
Ratnaningrum 2020 ⁑⁑	11 (15.9%)	–	–	–	–	–	8 (72.7%)	–	–	–	–	3 (27.3%)		1
SCORING = 2														
Illknur et al. 2009 ⁑	–	–	–	–	–	–	–	–	–	–	–	Archaeological sample: 1 (6.25%) Contemporary sample: 1 (3.5%)		–
Murlimanju et al. 2011 ⁂	57 (73.1%)	–	1 (1.3%)	1 (1.3%)	–	–	44 (56.4%)	0	–	Σ: 14 (17.9%)		Σ: 9 (11.%)		–
Walulkar et al. 2012 ⁂	♂: 63 (39.1%) ♀: 14 (21.9%) Σ: 77 (34.2%)	–	♂: 1 (1.3%) ♀: 0 Σ: 1 (1.3%)	♂: 4 (5.2%)♀: 0 Σ: 4 (5.2%)	–	–	♂: 45 (71.4%) ♀: 12 (85.7%) Σ: 57 (74.2%)	0	♂: 7 (11.1%) ♀: 1 (7.1%) Σ: 8 (10.4%)	♂: 3 ♀: 1 Σ: 7 (9.1%)	♂: 2 ♀: 2	–	–	–
Sudha et al. 2013 ⁂	–	–	–	–	–	–	–	–	–	13 (8.6%) Σ: 23 (7.6%)	10 (6.6%)	21 (14%) Σ: 34 (11.3%)	13 (8%)	–

Eboh and Obar-oefe 2014 ■	–	6 (6%)	–	–	–	–	–	–	–	–	–	4 (8%) Σ: 6 (6%)	2 (4%)	–
Ghosh et al. 2017	54 (45%)	165	2.4%	5.4%	10.9%		53.3%	0.6%	21.2%	–		–		–
Tallapaneni and Niveditha 2016 ⁂	59 (53.1%)	–	0	1 (0.9%)	3 Σ: 5 (4.5%)	2	R: 25 L: 34 Σ: 59 (53.1%)	1 (0.9%)	9 (8.1%)	1 Σ: 3 (2.7%)	2	2 Σ: 2 (1.8%)	0	–
Uday and Ratna Prabha 2016 ⁑⁑	113 (56.5%)	–	0	0	0	5 (4.4%)	R: 21 L: 43 Σ: 64 (56.6%)	0	52 (46%)	18 Σ: 46 (40.7%)	28	1 Σ: 6 (5.3%)	5	–
Ukoha et al. 2013 ⁂	–	–	–	–	–	–	–	–	–	–	–	3.6%	3.6%	–
Kalthur et al. 2017 *	–	–	–	–	–	–	–	–	–	–	–	R: 24% L: 10% ♂: 18.9% ♀: 11.6%		–
Lekshmy et al. 2017 ⁂	123 (61.5%)	–	R: 36 L: 30 Σ: 66 (53.6%)	34 (27.6%)	26 Σ: 56 (45.5%)	30	R: 58 R: 54 Σ: 112 (91%)	3 (2.4%)	29 (23.6%)	50 Σ: 98 (79.7%)	48	9 Σ: 20 (16.3%)	11	–
Nagarajan and Ganesh 2017 ⁂	–	–	–	–	–	–	–	–	–	–	–	–	–	single: 7 (87.5%) double:1 (12.5%) Σ: 8 (13.3%)

Article	Presence	Total no.	WB in main sutures						WB in specific sites of the skull					
			C	S	Sq-R	Sq-L	L	B	LL	A-R	A-L	P-R	P-L	I
			R: 3 L: 2		2	0	R: 23 L: 19			0	0	2	4	–
Narayan et al. 2019 ⁑, ⁂	13 (43%)	82	+ 4 bi-lateral = Σ: 9 (10.6%)	0	Σ: 2 (2.3%)		+ 10 bilateral =Σ: 52 (61.2%)	0	7 (8.2%)	+ 2 bilateral = Σ: 2 (2.3%)		Σ: 6 (7%)		–
Nayak and Shetty 2019 ⁂	24 (88.8%)	–	2 (7.4%)	–	–	–	6 (22.2%)	–	4 (14.8%)	18 (66.6%)		2 (7.4%)		–
Yadav and Salam 2020 ⁂	♂: 11 (30.6%) ♀: 8 (28.6%) Σ: 19 (29.7%)	–	1 (5.3%)	1 (5.3%)	1 (5.3%)		15 (78.9%)	–	4 (21%)	8 (42.1%)		–	–	–
SCORING = 3														
Pal and Routal 1986 ⁂	–	–	*D*:0 *M*: 0 *B*:0	*D*: 4 (5.8%) *M*: 0 *B:0*	*D*: 11 (8%) *M*: 1 (1.3%) *B*: 1 (5%)		*D*: 51 37%) *M*: 23 (30.3%) *B*: 9 (45%)	*D*: 0 *M*: 0 *B*: 0	*D*: 13 (18.8%) *M*: 4 (10.5%) *B*: 0	*D*: 8 (5.8%) *M*: 2 (2.6%) *B*: 2 (10%)		*D*: 8 (5.8%) *M*: 3 (3.9%) *B*: 2 (10%)		–
Gümüsburun et al. 1997 *	–	–	♂: 4 (2.1%) ♀: 5 4,5%) Σ: 9 (3%)	♂: 1 (0.5%) ♀: 4 (3.6%) Σ: 5 (1.6%)	♂: 14 (7.3%) ♀: 10 (9.1%) Σ: 24 (7.9%)		♂: 117 (60.9%) ♀: 70 (63.6) Σ: 187 (61.9%)	♂: 2 (1%) ♀: 0 Σ: 2 (0.7%)	♂: 26 (13.5%) ♀: 7 (3.6%) Σ: 33 (10.9%)	♂: 32 (16.7%) ♀: 14 (12.7%) Σ: 46 (15.2%)		♂: 22 (11.5%) ♀: 8 (7.3%) Σ: 30 (9.9%)		–
Govsa et al. 2014 ⁂	27	–	–	–	–	–	12 (4%)	–	–	–	–	–	–	*PI*: single:6 multiple: 3 Σ: 9 (3%) *II:* single: 2 multiple:4 Σ: 6 (2%)

Oguz et al. 2004 ⁂	–	–	–	–	–	–	–	–	–	–	–	♂: 0	♂: 4%	–
Cirpan et al. 2014 ⁂	–	–	–	–	–	–	–	–	–	–	–	–	–	3 (2%)
Cirpan et al. 2016 ⁑	89 (59.3%)	207	R:5 (3.3%) L:5 (3.3%)	7 (4.7%)	3 (2%)	10 (6.7%)	R: 56 (37.3%) L: 61 (40.7%)	3 (2%)	10 (6.7%)	10 (6.7%)	11 (7.3%)	8 (5.3%)	12 (8%)	–
Durge 2016	♂: 30 (41.7%) ♀: 42 (47.7%) Σ: 72 (45%)	–	♂: 4 ♀: 0	–	–	–	♂: 22 ♀: 34	♂: 2 ♀: 0	–	♂: 2 ♀: 0		♂: 0 ♀: 4		–
Basnet et al. 2019 ⁂	62 (88.6%)	–	3 (4.3%)	5 (7.1%)	29 (41.4%)		43 (61.4%)	0	8 (11.4%)	17 (24.3%)		18 (25.7%)		–
de Lucena et al. 2019 ⁂	–	–	–	–	–	–	–	–	–	♂: 6.7% ♀: 8.3% Σ: 19 (31.7%)	♂: 11.7% ♀:5%	–	–	–
Li et al. 2022 ⁂	182 (63.9%)	–	–	9 (4.9%)	–	–	143 (78.6%)	–	15 (8.3%)	22 (12.1%)		63 (34.6%)		7 (3.8%)
Murrieta-Angulo et al. 2019 ⁂■	–	–	–	–	–	–	–	–	–	–	–	0	0	–
Shiv et al. 2020 ⁂	♂: 19 (22.1%) ♀: 8 (18.2%) Σ: 27 (20.1%)	–	1 (0.8%)	7 (5.4%)	–	–	19 (14.6%)	–	–	–	–	–	–	–
SCORING = 4														
Finkel 1975 ⁂	♂: 114 ♀: 81	–	–	–	–	–	–	–	–	–	–	–	–	–
Goyal et al. 2019 ⁂	♂: 35 (36.1%) ♀: 17 (34%) Σ: 52 (35.3%)	–	9 (6.1%)	7 (4.8%)	–	–	41 (27.9%)	0	28 (19%)	3 (2%)		1 (0.7%)		–

Article	Presence	Total no.	WB in main sutures					WB in specific sites of the skull						
			C	S	Sq-R	Sq-L	L	B	LL	A-R	A-L	P-R	P-L	I
Marathe et al. 2010 ⁂	–	–	–	–	–	–	–	–	–	–	–	–	–	♂: 3 (1.4%) ♀: 2 (1.2%) Σ: 5 (1.3%)
Natsis et al. 2019 ⁂, ⁑⁑	♂: 67 (72.8%) ♀: 57 (77%) Σ: 124 (74.7%)	–	♂: 39 (42.4%) ♀: 27 (36.5%) Σ: 66 (39.8%)	♂: 9 (9.8%) ♀: 9 (12.2%) Σ: 18 (10.8%)	♂: 7 ♀: 5 Σ: 12	♂: 4 ♀: 5 Σ: 9	♂: 40 (43.5%) ♀: 34 (45.9%) Σ: 74 (44.6%)	♂: 2 (2.2%) ♀: – Σ: 2 (1.6%)	♂: 1 (1.1%) ♀: 2 (2.8%) Σ: 3 (2.4%)	♂: 15 ♀: 4 Σ: 19	♂: 17 ♀: 9 Σ: 26	♂: 2 ♀: 4 Σ: 6	♂: 2 ♀: 2 Σ: 4	–
Purohit and Yadav 2019 ⁑⁑	♂: 90 (72.6%) ♀: 41 (73.2%) Σ: 131 (72.3%)	–	♂: 10 (8.9%) ♀: 6 (14.6%) Σ: 14 (10.7%)	♂: 3 (3.3%) ♀: 5 (12.2%) Σ: 8 (6.1%)	Parieto-temporal: ♂: 12 (13.2%) ♀: 11 (26.8 %) Σ: 23 (17.6%) Parieto-mastoid: ♂: 7 (7.8%) ♀: 2 (4.9%) Σ: 9 (6.9%)		♂: 55 (61.1%) ♀: 32 (78%) Σ: 87 (66.4%)	0	♂: 24 (26.7%) ♀: 8 (19.5%) Σ: 32 (24.4%)	♂: 34 (37.8%) ♀: 15 (36.6%) Σ: (49) 37.4%		♂: 24 (26.7%) ♀: 13 (31.7%) Σ: 37 (28.2%)		–

* Percentage of WB (total number of bones) in specific sites calculated in relation to whole research sample (divided into sex groups, if distinguished)

⁑ Percentage of WB (total number of bones) in specific sites calculated in relation to group of individuals with present WB only (divided into sex groups, if distinguished)

⁂ Percentage of number of skulls that have WB in specific sites, in relation to whole research sample (divided into sex groups, if distinguished)

⁑⁑ Percentage of number of skulls that have WB in specific sites, in relation to group of individuals with present WB only (divided into sex groups, if distinguished)

■ Both sides of skull treated as an independent record (total number doubled)

Abbreviations: Presence – number and/or percentage of individuals with Wormian bones in sample. Total no. – total sum of Wormian bones in the whole sample. C – coronal suture, S – sagittal suture, Sq-R – right squamous suture, Sq-L – left squamous suture, L – lambdoid suture, B – bregma – LL – lambda, A-R – right asterion, A-L – left asterion, P-R – right pterion, P-L – left pterion, I – Inca bone [*II* – interparietal, *PI* – preinterparietal]. Variants of skull morphology: *D* – dolichocephalic, *M* – mesocephalic, *B* – brachycephalic. Parietal notch bones and ossicles in parieto-temporal or parieto-mastoid suture were considered as squamous sutural bones.

Table 3. Wormian Bones Prevalence in Male and Female Skulls (the articles using 3rd computational strategy selected only)

Article	WB examined in	Males	Females	Statistical significance
Finkel 1975	whole skull	114 (32.2%)	81 (29.6%)	not described
Marathe et al. 2010	Inca	3 (1.428%)	2 (1.176%)	not described
Walulkar et al. 2012	whole skull	63 (9.13%)	14 (21.87%)	not described
Singh 2012	asterion	– (82.14%)	– (85.19%)	not described
Masih et al. 2013	whole skull	– (4.1%)	– (3.6%)	not described
Natsis et al. 2019	whole skull	67 (72.8%)	57 (77%)	no significant difference
Yadav and Salam 2020	whole skull	11 (30.56%)	8 (28.57%)	not described
De Lucena et al. 2019	asterion	– (18.34%)	– (13.34%)	not described
Goyal et al. 2019	whole skull	35 (36.08%)	17 (34%)	no significant difference
Shiv et al. 2020	whole skull	19 (22.09%)	8 (18.18%)	not described
Yadav and Salam 2020	whole skull	11 (30.56%)	8 (28.57%)	not described