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Wormian bone types: investigating their appearance, correlation to sex, population affinity, and clinical syndromes

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Abstract

Background The types of Wormian bones may play a role in population affinity and differential diagnosis of several clinical syndromes. This study investigates the distribution of types in adult skulls, their correlation to sex, population affinity, and several impairments based on the literature. One hundred ten adult Turkish skulls, 80 (72.7%) males and 30 (27.2%) females, were investigated according to type, frequency, location, and sex. Horizontal and vertical diameters and distances from the mastoid process (MP) were measured with a digital caliper. SPSS 25 was used for all statistical analyses.

Results A total of 58 (52.72%) Wormian bones were identified from 110 skulls, 38 (65.5%) males and 20 (34.5%) females. The types of Wormian bones revealed significant differences between being on the right, left, or center ($p = 0.012$). The most frequent type was type 6 ($n = 14$, 24.1%) in males and type 7 ($n = 8$, 13.8%) in females. The most frequent type was type 7 in the lambdoid suture and type 8 in the parietomastoid suture.

Conclusions The present study revealed significant differences regarding the asymmetric distribution of Wormian bones and unilateral asymmetrical types in Turkish skulls. Several factors could contribute to this, including underlying clinical syndrome, deficiencies in embryological development, and population affinity.

Keywords Wormian bones, Os incae, Lambdoid suture, Lambda, Skull

Background

The squamous part of the occipital bone is sometimes divided by one or more transverse sutures at the level of the superior nuchal line. The part above the transverse suture is called the Wormian bone (os incae). The Wormian bone was named after the Dutch anatomist Olaus Wormius (1588–1654) (Porter 1963); Thomas Bartholin coined the term "Wormian bone" in honor of Wormius (Bellary et al. 2013). The same variation was called the interparietal or sutural bone, as it occurs along the sutures (Hanihara and Ishida 2001). The presence of longitudinal or transverse sutures causes the formation of more than one Wormian bone. According to the literature, these accessory bones are seen unilaterally, mainly on the right side, and frequently in the lambdoid suture (LS), coronal suture (CS), and Lambda (Bellary et al. 2013; Sanchez-Lara et al. 2007). Other locations include Bregma, Pterion, sagittal suture (SS), occipitomastoid suture (OMS), parietomastoid suture (PMS) (Showri and Suma 2016), and Asterion (Bellary et al. 2013; Sanchez-Lara et al. 2007). Several theories have been proposed regarding the reasons for the development of Wormian bones, but the mechanism behind the formation of this accessory bone has not been clarified yet (Goyal et al. 2019). Some authors have reported that these bones are formed through embryological development or genetic

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influences, while others have reported that they develop as a result of external influences (Goyal et al. 2019). Bellary et al. reported that Wormian bones developed from secondary ossification centers (Bellary et al. 2013). Matsumura et al. reported that Wormian bone formation was caused by a lack of interparietal bone fusion, the formation of pre-interparietal bone, and multiple ossification centers along the sutures (Matsumura et al. 1993). In other studies, increased intracranial pressure, sutural width, stress due to mechanical deformation, early fusion of the sutures, various metabolic bone disorders, and incomplete ossification have been hypothesized to cause Wormian bone formation (Bellary et al. 2013; Ossenberg 1976). Some studies have reported a relationship between the incidence and distribution of Wormian bones and cranial deformation (Lahr 1996; Ossenberg 1976). Therefore, the aim of this study was to investigate the Wormian bone types in Turkish skulls according to sex and direction. Another aim of this study was to examine the relationship between Wormian bone types and diseases, based on the literature.

Methods

This study was performed on 110 adult Turkish skulls. The skulls were provided by the Akdeniz University, Faculty of Medicine, Department of Anatomy, Antalya, Turkey. The study was approved by the Akdeniz University School of Medicine Ethics Committee and is in line with the 1964 Declaration of Helsinki, approval date August 26, 2020, and number 614. The classification of Kadanoff and Mutafov was used to determine Wormian bone types due to the different criteria and terminology (Kadanoff and Mutafov 1964). Complete ossified skulls without any pathology were included in this study. Twenty-one skulls with fractures, cranial deformation, and trauma were not included in the study. To achieve intraobserver precision, three widely used precision estimates were calculated: the technical error of measurement (TEM), the relative technical error of measurement (rTEM), and the coefficient of reliability (R) (Akdag et al. 2020; Guzelad et al. 2023; Ogut et al. 2022, 2021; Ogut and Yildirim 2021; Ögüt et al. 2022; Regoli et al. 2016; Sekerci et al. 2021). The possible relationship between clinical syndromes and Wormian bones was investigated through a review of the published literature. All measurements were taken using a digital caliper (0.01–1000 mm, Mitutoyo, Japan). The following parameters were explored.

1. The incidence of Wormian bones
2. The location of Wormian bones
3. The types of Wormian bones
4. The size of Wormian bones
5. The distance between the mastoid process and Wormian bones (MP-W)

Statistical analysis

Descriptive statistics (mean, minimum, maximum, standard deviation) were performed on all parameters. Differences between the data of skull measurements were analyzed by the Student's *t* test. The sex differences in the study were analyzed by the chi-square test. SPSS 25 (IBM, USA) was used for all statistical analyses. $p < 0.05$ was accepted as a significant difference.

Results

Technical error of measurement (TEM) values of all variables measured were 0.01–0.13 mm, and relative technical error of measurement (rTEM) was 0.12–4.38%. The R values of most variables were close to 1, suggesting that an acceptable degree of intra-observer precision was obtained for the measurements.

Incidence and location of Wormian bones

Wormian bones were detected in 58 (52.72%) of 110 Turkish skulls. This rate was 20 (34.5%) in females and 38 (65.5%) in males (Fig. 1). However, there was no difference between the sexes. There were 16 (27.16%) Wormian bones in the center, 22 (37.9%) on the right, and 20 (34.5%) on the left (Table 1). In addition, 25 Wormian bones were found in the LS (43.1%), 18 in the PMS (31%), 7 in the OMS (12.1%), 6 in the Lambda (10.3%), and 2 in the SS (3.4%). There was a statistically significant difference in the presence of Wormian bones on the right, left, or center ($X^2 = 19{,}59$; $p = 0.012^*$) (Table 1). This revealed an asymmetric distribution.

The types of Wormian bones

The detected Wormian bone shapes were recorded as oval, triangular, quadrangular, and irregular, and seven types of Wormian bones were identified in the current study. The detected Wormian bone types are shown in Fig. 2. The classification of Kadanoff and Mutafov (1964) was used for reporting the types (types 1–12) (Kadanoff and Mutafov 1964). The recorded types were os incae totum (type 1), os incae bipartite (type 2), os incae quadripartite (type 4), os incae laterale sinistrum (type 6), os incae centrale (medium) (type 7), os incae laterale dextrum (type 8), and os incae duplex symmetrical (type 9) (Fig. 2). Os incae centrale (medium) (type 7) was more frequent in females and os incae laterale sinistrum (type 6) in males (Table 2). Type 3 (Os incae tripartite), type 5 (os incae multipartite), type 10 (os incae duplex asymmetrical), type 11 (pars incoidea squamae occipitalis; processus sagittal squamae occipitalis), and

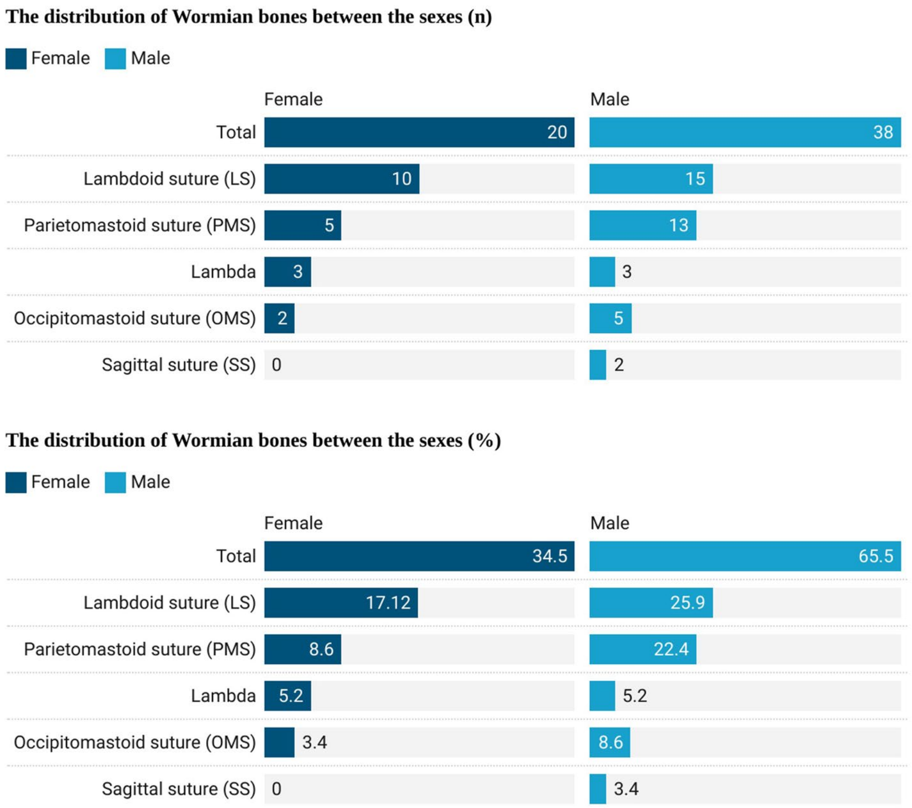

Fig. 1 The distribution of Wormian bones between the sexes (*n*/%). Wormian bones were found in 10 (%17.12; female) in 15 (%25.9; male) in Lambdoid suture (LS), 3 (%5.2) in Lambda in both sexes, 5 (%8.6; female) in 13 (%22.4; male) in parietomastoid suture (PMS), 2 (%3.4; male) in Sagittal suture (SS), and 5 (%8.6; female) in occipitomastoid suture. A total of 58 (52.72%) Wormian bones were identified from 110 skulls, 38 (65.5%) male and 20 (34.5%) female

Table 1 Symmetry/asymmetry analysis of Wormian bones (*n*/%)

Location	**Direction**			
	Center	**Right**	**Left**	**Total**
Lambdoid suture (LS)	9 (%15.5)	7 (%12.1)	9 (%15.5)	**25 (%43.1)**
Lambda	4 (% 6.9)	1 (% 1.7)	1 (% 1.7)	**6 (%10.3)**
Parietomastoid suture (PMS)	1 (% 1.7)	9 (%15.5)	8 (%13.8)	**18 (%31)**
Sagittal suture (SS)	2 (%3.4)	–-	–-	**2 (%3.4)**
Occipitomastoid suture (OMS)	–-	5 (% 8.6)	2 (%3.4)	**7 (%12.1)**
Total	16 (%27.16)	22 (%37.9)	20 (%34.5)	**58 (%100)**

Chi-square tests $X^2 = 19{,}59$; $p = 0.012$, $p<0.05$ (direction-location)

(See figure on next page.)

Fig. 2 The recorded Wormian bone types (posterior view of the skull). **a** os incae totum (Type 1) describes the complete undivided type. **b** os incae bipartite (Type 2) describes the two-part Wormian bone. **c** os incae quadripartite (Type 4) describes the four-part Wormian bone. **d** os incae laterale sinistrum (Type 6) shows the unilateral Wormian bone on the left side of the skull. **e** os incae centrale (medium) (Type 7) shows a single Wormian bone. **f** os incae laterale dextrum (Type 8) shows the unilateral Wormian bone on the right side of the skull. **g–h** os incae duplex symmetrical (Type 9) shows bilateral Wormian bones. The numbers on the figure indicate the number of Wormian bones

Fig. 2 (See legend on previous page.)

Table 2 Evaluation of Wormian bone types according to sex, location, and direction (*n*/%)

	Type 1	Type 2	Type 4	Type 6	Type 7	Type 8	Type 9	Total
Female	1 (%1.7)	--	1 (% 1.7)	2 (%3.4)	8 (%13.8)	5 (% 8.6)	3 (%5.2)	20 (%34.5)
Male	--	1 (% 1.7)	--	14 (%24.1)	9 (%15.5)	12 (%20.7)	2 (%3.4)	38 (%65.5)
Total (sex)	1 (%1.7)	1 (%1.7)	1 (%1.7)	16 (%27.6)	17 (%29.3)	17 (%29.3)	5 (% 8.6)	58 (%100)
Lambdoid suture (LS)	--	--	1 (% 1.7)	7 (%12.1)	9 (%15.5)	5 (% 8.6)	3 (%5.2)	25 (%43.1)
Lambda (La)	--	--	--	1 (% 1.7)	4 (% 6.9)	--	1 (% 1.7)	6 (%10.3)
Parietomastoid suture (PMS)	1 (%1.7)	1 (%1.7)	--	6 (%10.3)	2 (%3.4)	8 (%13.8)	--	18 (%31.0)
Sagittal suture (SS)	--	--	--	--	1 (%1.7)	1 (%1.7)	--	2 (%3.4)
Occipitomastoid suture (OMS)	--	--	--	2 (%3.4)	1 (%1.7)	3 (%5.2)	1 (%1.7)	7 (%12.1)
Total (location)	1 (%1.7)	1 (%1.7)	1 (%1.7)	16 (%27.6)	17 (%29.3)	17 (%29.3)	5 (% 8.6)	58 (%100)
Center	--	--	--	--	15 (%25.9)	1 (%1.7)	--	16 (%27.6)
Right	1 (%1.7)	--	--	1 (%1.7)	1 (%1.7)	16 (%27.6)	3 (%5.2)	22 (%37.9)
Left	--	1 (%1.7)	1 (%1.7)	15 (%25.9)	1 (%1.7)	--	2 (%3.4)	20 (%34.5)
Total (direction)	1 (%1.7)	1 (%1.7)	1 (%1.7)	16 (%27.6)	17 (%29.3)	17 (%29.3)	5 (% 8.6)	58 (%100)

Chi-square tests $X^2 = 86{,}665$; $p = 0.001$, $p<0.05$ (direction-type), $X^2 = 10{,}573$; $p = 0.102$ (sex-type)

type 12 (mendosal suture, MS) were not detected in the present study. Type 7 was most frequently detected in LS ($n = 9$, 15.5%), while type 8 was most frequent in PMS ($n = 8$, 13.8%) according to the location. The types of Wormian bones revealed significant differences according to their presence on the right, left, and center ($X^2 = 86{,}665$; $p = 0.001$, $p<0.05$). However, the detected types did not show any differences according to sex ($X^2 = 10{,}573$; $p = 0.102$) (Table 2).

The size and the distance between the mastoid process and Wormian bones (MP-W)

The mean vertical diameters (VD) of Wormian bones were 18.24 ± 4.14 mm (female) and 14.84 ± 9.41 mm (male), and the mean transverse diameters (TD) were 13.24 ± 6.67 mm (female) and 16.90 ± 16.33 mm (male). Although Wormian bones in females were longer than in males, there were no differences in their mean VD ($p = 0.131$). The TD of the Wormian bones in males was bigger than in females. However, there were no differences between the diameters according to sex ($p = 0.343$). The mean distances to the MP were 1.74 ± 0.42 mm (female) and 1.74 ± 0.47 mm (male) ($p = 0.952$) (Table 3).

Discussion

Incidence and location

In the present study, 25 Wormian bones were found in the LS (43.1%), 18 in the PMS (31%), 7 in the OMS (12.1%), 6 in the Lambda (10.3%), and 2 in the SS (3.4%) in Turkish skulls. There was a difference in the presence of Wormian bones on the right, left, or center. Thus, the current study revealed an asymmetric distribution in Turkish skulls. It has been reported that Wormian bones were found with an incidence of 56.06% ($n = 132$) in LS (45.45%), Lambda (13.63%), PMS (8.33%), CS (6.06%), Asterion (5.30%), SS (4.54%), OMS (2.27%), PSS (2.27%), and Pterion (1.51%) in Indians (Showri and Suma 2016). Similarly, in many studies, Wormian bones were frequently observed along the LS in Indian, Turkish, and Greek adult skulls (Albay et al. 2013; Goyal et al. 2019; Kiliç-Safak et al. 2020; Murlimanju et al. 2011; Natsis et al. 2019). Goyal et al. found this variant (35.3%, $n = 52$) in LS (27.89%), CS (6.12%), SS (4.76%), Asterion (2.04%), Lambda (19.04%), and Pterion (0.68%) in North Indians (Goyal et al. 2019). These divergent bones were more frequently found in left LS and less in right OMS in Anatolian populations (Cirpan et al. 2015). In a study (Govsa et al. 2014), Wormian bones ($n = 27$) were

Table 3 The vertical diameter (VD), transverse diameter (TD), and distance between the mastoid process and Wormian bones (MP-W)

The size and the distance between the MP-W (mm)	Female (mean ± SD)	Male (mean ± SD)	*F*	*t*	df	*p*
VD	18.24 ± 4.14	14.84 ± 9.41	7.060	− 1.534	56	**0.131**
TD	13.24 ± 6.67	16.90 ± 16.33	4.012	0.956	56	**0.343**
MP-W	1.74 ± 0.42	1.74 ± 0.47	3.78	0.060	56	**0.952**

Student's t test (*MP* mastoid process, *TD* transverse diameter, *VD* vertical diameter, *W* Wormian, *MP-W* the distance between the mastoid process and Wormian bone, *SD* standard deviation) detected as interparietal (2%), preinterparietal (3%), and sutural bones (4%) in the LS in the 300 adult skulls. The LS (44.6%), followed by the CS (39.8%), Asterion (21% on the left and 15.3% on the right), and PMS (15.1% on the left and 13.3% on the right) in Greek adult skulls. Other sutures that displayed Wormian bones were the squamosal, zygomaticosphenoid, metopic, frontonasal, and frontozygomatic sutures in Greek adult skulls (Natsis et al. 2019). In a study on 50 skulls, Wormian bone was found in 28% in the LS, 2% in the CS, 8% in Lambda, 4.3% in Bregma, and 8% in squamous part and metopic sutures in Turkish skulls (Albay et al. 2013). In another study on 166 Greek adult skulls, it was found that 74.7% of skulls ($n=124$) displayed Wormian bones, and there were no differences between the incidence of Wormian bones by sex and age. Wormian bones (59.3%) were found in the left LS (40.7%) and the right OMS (1.3%) in 150 skulls in the West Anatolian population (Cirpan et al. 2015). They have been reported as 18% in Asterion and 2% in OMS in Turkish skulls (Albay et al. 2013). In another study, the highest occurrence was recorded at the LS (53.33%), while the lowest incidence was reported at the Bregma and metopic sutures (0.61%) in 120 East Indian skulls (Ghosh et al. 2017). In a study conducted in Nigeria, 22 skulls were analyzed, and the area with the highest prevalence of Wormian bone (45.46%) was the LS (Uchewa et al. 2018). The findings of a retrospective French study that used 605 CT scans from a child population reported that the prevalence of Wormian bones (53%) was detected in the LS (Marti et al. 2013). Wormian bones were found in the coronal, squamosal, and SS in six of 25 Malaysian skulls, although they were frequently found in the LS and fontanelles (Khan et al. 2011). The Bregmatic type of Wormian bone has been reported to be associated with Wormian bones in the SS and LS in Caucasians (Barberini et al. 2008). Wormian bones were found to be more common in LS in Turkish skulls, which is consistent with the literature. However, it has been noted that Turkish skulls have more asymmetrically located Wormian bones instead of the types located in the fontanelles, such as the Bregmatic type.

The authors use the mastoid process (MP) as a landmark for metric analyses because it is a palpable bony structure that enables the determination of the Wormian bones. The distance between the apex of the MP and the Wormian is a valuable parameter for anthropology, neurosurgery (for possible clinical syndromes), anatomy, and forensic sciences (Aydın Kabakçı et al. 2021). This study found the distance from the Turkish population's average value to identify the possible location of Wormian bones. The current study revealed that the mean distances between the MP and Wormian bones were approximately 1.74 ± 0.4 mm in Turkish skulls Based on this, in the present study, it can be said that the Wormian bones were located more posteriorly, and these accessory bones were close to the MP. Some groups of culturally deformed skulls also exhibited a higher frequency of apical, PMS, and OMS-located Wormian bones. Furthermore, the group with varying degrees of cultural modification displayed more Lambdoid Wormian bones than the group without deformation (O'Loughlin 2004). O'Loughlin implied that environmental pressures might have a greater impact on the formation of Wormian bone in posteriorly positioned sutures than in their anteriorly positioned counterparts in 127 deformed and undeformed skulls (O'Loughlin 2004). According to this theory, cranial deformation and variations in the population affinity might affect the location and incidence of Wormian bone types. Previous studies were presented in Table 4.

Types

In the present study, the classification of Kadanoff and Mutafov (1964) was used to identify the types of Wormian bones. The following types were found in the present study: types 1, 1 (1.7%); type 2, 1 (1.7%); type 4, 1 (1.7%); type 6, 16 (27.6%); type 7, 17 (29.3%); type 8, 17 (29.3%); and type 9, 5 (8.6%). These rates show that unilateral and asymmetrical types are more common in the Turkish population. In the present study, type 3 (os incae tripartitum), type 5 (os incae multipartitum), type 10 (os incae duplex asymmetricum), type 11 (pars incoidea squamae occipitalis; processus sagittalis squamae occipitalis), and type 12 (MS) were not detected. However, os incae laterale sinistrum (type 6) of unilateral asymmetrical types was identified with a rate of 27.6%, os incae centrale (medianum) (type 7), and os incae laterale dextrum (type 8) were detected with a rate of 29.3% in Turkish skulls. The detected types did not show any significant difference according to sex (Table 2). Hanihara et al. reported that type 1 and type 7 were seen more frequently than other types in several populations (Hanihara and Ishida 2001). Moreover, the prevalence of Wormian bones was often higher in New World populations than in northeast Asians and Australians. The populations of Tibetan/Nepalese and Assam/Sikkim in northeast India have a greater number of Wormian bones than those in surrounding areas. It has been reported in the literature that types 6, 8, and 10 are less frequent, and type 9 is rare. In a study (Govsa et al. 2014), it was reported that Wormian bones were divided into the following types: interparietal (2%), pre-parietal (3%), and sutural bones (4%) in 300 adult skulls. It was reported that the interparietal bones were rhomboid, the pre-interparietal ones were triangular, and the sutural bones were round or heart-shaped (Govsa et al. 2014). It has also been

Table 4 Previous studies based on the literature

Authors	**Year**	**Study design**	**Number (*n*)**	**Age**	**Wormian (%/*n*)**	**Location**	**Country**	**Side**	**Disorders**
Pryles et al	1979	CT	515	0–14	17%(91)	-LS -Lambda	USA (Brooklyn)	Unilateral/ bilateral	-CNS abnormalities (micro and macrocephaly, hydrocephalus, craniosynostosis, cerebral palsy, epilepsy, and learning difficulties) - Wormian bones are a marker for CNS developmental abnormalities
Hanihara et al	2001	Dry skull	> 8000	-	Types 1–6	-LS -MS -Lambda	Japan UK France USA Russia	Unilateral/ bilateral	- Artificial cranial deformation - Secondary trait formed by ontogenetic stress
Barberini et al	2008	3D CT	one	66 years	1 (Pentagonal bregmatic type)	-Bregma	Italy	Unilateral	-Its formation at the neural crest–mesoderm -It represents an additional ossification center
Wu et al (2011)	2011	3D CT	210	0–2	n=11	-MS	USA	Unilateral/ bilateral	- Craniosynostosis (coronal and metopic sutures) - Scaphocephaly, craniofacial anomaly, FGFR3,pro250arg mutation (Muenke syndrome)
Stotland et al	2012	Intraoperative	one	2,5	1 Pentagonal bregmatic type	-Bregma	USA	Bilateral	-Metopic synostosis, trigonocephaly
Marti et al	2013	CT	605	< 3 months 3–6 months 6–12 months 1–2 years 2–3 years	53% (320) − 1–3: 43% (260) ≥ 4: 10% (60) ≥ 5: 6% (40)	-SS (22) -LS (252, right; 256, left) -PSS (45 right; 41 left) -PF (178) -PLF -MF	France	Unilateral/ bilateral	-Group A: headaches, convulsive seizures, neurological deficits (*n* = 349; 445 Wormian) -Group B: head injuries (*n* = 132; 180 Wormian) -Group C: hydrocephalus (*n* = 55; 73 Wormian) -Group D: plagiocephaly (12) and craniosynostosis and cranial anomaly (*n* = 69; 96 Wormian) -OI (10%) differential diagnosis
Thanapaisal et al	2013	Dry skull	400	16–93	7.25%(29) 11 types	-Types 1–3 -Types 8–10	Thailand	Unilateral/ bilateral	It can be used in the differential diagnosis of the fracture line and personal identification
Showri et al	2016	Dry skull	132	-	56.06% Rectangular (52.7%) Irregular (37.83%) Triangle (9.45%)	-LS (45.45%) -Lambda (13.63%) -PMS (8.33%) -CS (6.06%) -Asterion (5.30%) -SS (4.54%) -OMS (2.27%) -PSS (2.27%) -Pterion (1.51%)	India (Bengaluru)	Unilateral/ bilateral	-MSX2 gene variation -Hydrocephalus -Some cerebral diseases -Artificial cranial deformation

Table 4 (continued)

Authors	Year	Study design	Number (*n*)	Age	Wormian (%/*n*)	Location	Country	Side	Disorders
Goyal et al	2019	Cadaver	147	Adult	35.3%(52)	-LS:41 (27.89) -CS:9 (6.12) -SS:7 (4.76) -Asterion 3 (2.04) -La:28 (19.04) -Pterion: 1 (0.68)	North India (Haryana)	Unilateral/ bilateral	-The diagnosis and management of Wormian-related diseases -The detection of physical abuse -Fracture line diagnosis
Present study	2022	Dry skull	110	Adult	52.72 %(58) 7 types	-LS 25 (%43.1) -Lambda 6 (%10.3) -SS 2 (%3.4) -PMS 18 (%31.0) -OMS 7 (%12.1)	Turkey	Unilateral/ bilateral	- Cranial anomalies - The indicator of clinical syndromes and population affinity

reported that interparietal and preparietal bones were single, and sutural bones were multiple (Govsa et al. 2014). This might be due to the differences in the ossification process of the Wormian bone during embryological development. In the present study, the high rate of the asymmetrical distribution of Wormian types in Turkish skulls might be due to the common congenital background of populations. It has been claimed that unilateral asymmetrical types were formed due to the failure of embryological development, impairment in the ossification centers, or congenital disorders (Goyal et al. 2019; Stotland et al. 2012). It has also been reported that symmetrical Wormian bones were reported in 12.5% of South Indian skulls (Ghosh et al. 2017) and that genetic factors primarily influence the unilateral/bilateral presence of Wormian bones (Ghosh et al. 2017). The asymmetric types in this study confirm the correlation between Wormian types and population affinity.

Population affinity

Population affinity seems to affect the incidence of Wormian bones, which may indicate genetic influences on the formation of Wormian bones (Barberini et al. 2008). Therefore, the distribution of the Wormian bone frequency suggests a possible congenital background for forming this bone (Hanihara and Ishida 2001). In the present study, the different frequencies of 110 adult Turkish skulls show that the incidence is similar to the Indian and European populations, but there may be regional differences. It is usually observed as an anatomical variant in Asian populations but may be related to pathology in European populations (Brothwell 1959). The location of the Wormian has population-specific variations. For instance, Chinese populations displayed the highest incidence of Wormian bones (80%) compared with different populations (Brothwell 1959). There were also significant differences between populations in different parts of India. In a study, 57 skulls (73.1%) displayed Wormian bones in South India, while this rate was 35.57% in North India in 147 skulls of the Haryana population. It has been observed that there was a significant difference between regions, and the incidence was significantly higher in South India compared to other regions of India (Goyal et al. 2019).

Although a lower frequency of Wormian bone has been detected in Northeast Asians and Australians, it has been reported that it is frequently seen in Tibet, Nepal, and some African populations (Hanihara and Ishida 2001). In Anglo-Saxons, this rate is 55%. The incidence of Wormian bone is lower in Central and Western Asia and Europe (Hanihara and Ishida 2001). Previous studies indicated a significant frequency of Wormian bones (74.7%) in Greek populations (Natsis et al. 2019). In a study, 59.3% of skulls ($n = 150$) were found to have Wormian bones in the West Anatolian population (Cirpan et al. 2015). This incidence rate is lower than in previous studies, which may be due to differences in population affinity (Cirpan et al. 2015). This bone is more prevalent in Asians than Europeans, and asymmetrical variabilities are more prevalent in Anatolia (including Turkish skulls) and Asia, suggesting a hereditary distribution.

Sex

In the present study, males ($n = 38$; 65.5%) displayed more Wormian bones than females ($n = 20$; 34.5%), but there was no significant difference according to sex. The most frequent types were type 7 ($n = 8$, 13.8%) in females and type 6 ($n = 14$, 24.1%) in males in Turkish skulls. Therefore, in the present study, considering the relationship between the types, it can be said that Wormian bones are located unilaterally on the left side in males and more centrally in females. Some studies have claimed that males have more Wormian bones than females in northeastern Thailand populations (Thanapaisal et al. 2013);

other studies have reported that there were no significant differences between the sexes (Hanihara and Ishida 2001; Natsis et al. 2019; Zambrano et al. 2021). Goyal et al. found Wormian bones in 52 skulls (35.3%) out of 147 in Indians. This rate was 23.8% in males and 11.5% in females, and no significant difference was found according to sex (Goyal et al. 2019). Hanihara et al. reported no significant differences between the sexes in the formation and incidence of Wormian bones (Hanihara and Ishida 2001). Although central (medium) types were more frequent in females and unilateral in males, it can be said that the types have a similar distribution between both sexes.

Age

Most of the reviewed literature, including the present study, focuses on adult skulls. There seems to be no association between Wormian bone appearance and age groups in adult individuals; however, there is aliterature exploring that correlation for subadults. It has been reported that there was no relationship between the stress on the deformed skull and the formation of Wormian bone according to the age factor (Goyal et al. 2019). In the study of Pryles et al., which included 515 radiographs taken from children aged 0–14, they reported that the incidence and number of Wormian bones decreased with ageing, associated with the closure of the cranial sutures (Pryles and Khan 1979). This rate decreased from 68% in the 0–4 age group to 24% in the 5–9 age group and 8% in the 10–14 age group. Marti et al. found a lower frequency in the 0–3 age range (Marti et al. 2013). Wormian bone was also observed on ultrasonography of the fetus in the prenatal period (Goyal et al. 2019). However, a study reported that there were no differences in the incidence of Wormian bones according to age (Natsis et al. 2019). Although there are studies with different age groups in the literature, they are insufficient to establish a definite correlation between the presence of Wormian bones and the age factor.

Underlying pathologies and clinical syndromes

The clinical syndromes of the skulls were unknown in the current study, and therefore, the relationship between the syndromes and the Wormian bones was based on the literature. There are pathological and diagnostic implications for Wormian bones, such as quantity, arrangement, and size, which are significant in distinguishing these accessory bones from a pathology (Cremin et al. 1982). These variants might be significant markers of disorders that seem to be silent. Specific factors may be considered pathologically relevant before the existence of Wormian bones, such as the quantity of Wormian bone, their mosaic-like design and their dimensions (Cremin et al. 1982). Ghosh et al. stated that 2.5% of the skulls ($n = 120$) included at least ten Wormian bones, which is a strong indicator for pathology. Similarly, a study in the skull radiographs of 81 cases of osteogenesis imperfecta (OI) reported more than ten Wormian bones in OI patients in radiography (Cremin et al. 1982). A retrospective study of 195 patients reported that Wormian bones were detected in 35% of patients with OI type 1, 96% with OI type 3, and 78% with OI type 4 (Semler et al. 2010).

Several studies have asserted that these accessory bones were more frequently seen in pycnodysostosis, rickets (healing phase), kinky hair syndrome, Menke's syndrome, cleidocranial and craniometadiaphyseal dysplasia, congenital hypothyroidism, hypophosphatasia, otopalatodigital syndrome, Hadju Cheney (primary acro-osteolysis syndrome), Ritscher-Schinzel syndrome (craniofacial, cerebellar, and cardiac anomalies), Down's syndrome, mental acro-osteolysis, pachydermal retardation, congenital cutis laxa, and corpus callosum agenesis (Bellary et al. 2013; Kaplan et al. 1991; Papadopoulou et al. 2005; Sanchez-Lara et al. 2007; Semler et al. 2010; Stotland et al. 2012). Table 4 shows summaries of the previous studies that have been done on this subject. The fact that Wormian bones are more common in specific gene mutations, various diseases, and cases of cranial deformation suggests that these variants can be used in the differential diagnosis of several clinical syndromes.

In a case study of a 66-year-old Caucasian female, it was claimed that it is an essential regulatory center of cranial growth and may mediate interactions on which cranial development is dependent (Barberini et al. 2008). Craniosynostosis can occur when this border is lost due to gene mutation, indicating that the Wormian bone is tightly linked to brain development. Signals from the dura mater, such as fibroblast growth factors, can regulate the patency of the cranial sutures. Soft tissues, such as meningeal introgression of the cerebral falx, have an essential role in the neurocranial organization; therefore, they can affect bone morphology (Barberini et al. 2008). In disorders with impaired skull ossification, this results in larger cranial sutures and fontanelles and a prolonged period in which the skull remains pliable (Bellary et al. 2013; Sanchez-Lara et al. 2007). Similarly, a study which obtained from 20 deformed pre-Columbian skulls (10 from Mexico and 10 from Peru) and 20 normal skulls reported that Wormian bones were more frequent in the brachiocephalic skulls. Their presence may occur due to mechanical factors and dural pressures between the sutures (Sanchez-Lara et al. 2007). In a case study with a 14-week-old female with metopic synostosis and pentagonal bregmatic Wormian bone, it was claimed that there is a correlation between ossification deficiencies, skull asymmetry, endochondral bone malformation, metopism, and the formation of Wormian bones (Stotland et al. 2012). However, this correlation can not be made with certainty as the medical records of the deceased were not available to the researchers.

In a study of 515 infants and children, it was reported that Wormian bone is associated with abnormal development of the CNS, and it can be used as a marker for early diagnosis and treatment of clinical syndromes. It was reported that 91 (17%) of 515 infants and children had Wormian bones, 82 of these children (90%) had a primary CNS disorder, and five (6%) had minimal brain dysfunction syndrome (Pryles and Khan 1979). A retrospective analysis of 605 CT brain scans in children aged 0 to 3 years reported that 445 Wormian bones were identified in those with headache, convulsive seizures, neurological defects, and focal neurological signs, 73 Wormian bones in hydrocephalus, and 96 Wormian bones in those with cranial bone abnormalities, including plagiocephaly and craniosynostosis (Marti et al. 2013). Therefore, it can be said that Wormian bones may develop due to rapid enlargement of the cerebrum, increased cranial volume, greater intracranial pressure, and expansion of sutures, which may explain why they were found in greater numbers in patients with hydrocephalus and several cranial abnormalities (Marti et al. 2013). Therefore, in the present study, the different types of Wormian bones may also be associated with various clinical syndromes with craniofacial abnormalities.

Other theories put forward on this subject were as follows: In a microCT imaging study from 10 horses involving the biomechanical analysis of Wormian bone, it was reported that these bones acted as a protective mechanism to protect the brain from skull injury or trauma (Zambrano et al. 2021) and that the development of Wormian bone was related to natural selection rather than a functional adaptation of bone (Zambrano et al. 2021). A study conducted in a large geographical area reported that artificial cranial deformation and ontogenetic stress also affect the formation of Wormian bone (Hanihara and Ishida 2001). Another study of 300 adult skulls reported that this variation could help identify the exact manner and cause of death, murder, or child abuse, and the analysis of fatal injuries (Govsa et al. 2014). As emphasized in this study, the detection of Wormian bones may be an indicator of many syndromes or traumas.

Future directions

Understanding Wormian bone development may lead to a greater knowledge of other bony malformations that manifest as clinical disorders. Disturbances in the formation of Wormian bones may result in various craniomorphological disorders and clinical syndromes. In addition to that, Wormian bones are used as skeletal identifiers in diagnosing several congenital disorders; therefore, it can also aid in reconstructing the biological profile and positive identification of unknown remains. Observation of these extra bones in specific populations may potentially aid in the early detection of different disorders. This bone is more prevalent in Asians than Europeans, and asymmetrical variabilities are more prevalent in Anatolia and Asia, suggesting a hereditary distribution. For this reason, examining the genetic sequence of diseases with a diagnosis of Wormian bones and investigating them in more homogeneous and large-scale groups will strengthen their relationship with Wormian bones and clinical syndromes.

Limitations

The correlation between Wormian bone and neurodegenerative or psychiatric diseases such as Parkinson's, Alzheimer's, schizophrenia, anxiety, and depression has not been studied, and there is a deficiency in the literature on this subject. Imaging modalities were not employed in the present study. Patients with Wormian bones should receive 3D/4D radiological imaging to determine which types of Wormian bones are linked to certain diseases. Another idea would be to perform studies on anonymized CT or MRI data from hospitals for patients with a known medical history of clinical syndromes/diseases under study, and to perform a screening test for the presence of Wormian bones, to proceed and then calculate the incidence and correlation rates.

Conclusions

The present study revealed significant differences regarding the asymmetric distribution of Wormian bones and unilateral asymmetrical types in Turkish skulls. Several factors could contribute to this issue, including underlying clinical syndrome, deficiencies in embryological development, and population affinity.

Abbreviations

CNS	Central nervous system
CS	Coronal suture
CT	Computed tomography
FS	Frontal suture
LS	Lambdoid suture
MF	Mastoid fontanel
MP	Mastoid process
MP-W	The distance between the mastoid process and Wormian bone
MS	Mendosal suture
OI	Osteogenesis imperfecta
OMS	Occipitomastoid suture
PMS	Parietomastoid suture
PSS	Parietosquamous suture
R	Coefficient of reliability
rTEM	Relative technical error of measurement
SPSS	Statistical Package for the Social Sciences
SS	Sagittal suture
TEM	Technical error of measurement

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Authors' contributions

EO and FBY were involved in the conception and design of the study, acquisition of the data, analysis, and interpretation of the data. EO and FBY contributed to drafting the article and revising it critically for important intellectual content. EO and FBY participated in the final approval of the version to be submitted.

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Availability of data and materials

The dataset used and analyzed during the current study is available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

This study was approved by the Clinical Research Ethics Committee of Akdeniz University under the ethical standards in the 1964 Declaration of Helsinki on August 26, 2020, with protocol number 614. All procedures were executed strictly following the tenets of the Declaration of Helsinki.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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