BJCR BRAZILIAN JOURNAL OF CASE REPORTS

PCAA PUBLICAÇÕES CIENTÍFICAS DE ACESSO ABERTO

Case Report

Multiple Wormian Bones in the Lambdoid Suture: A Report of Rare Occurrence

José Aderval Aragão [1,*], Roberto Ferreira de Oliveira [2], Smilly Oliveira de Sousa Silva [3], Lucas Inacio Riffel [4], Iapunira Catarina Sant'Anna Aragão [5], Felipe Matheus Sant'Anna Aragão [6], Giulianna da Rocha Borges [7], Francisco Prado Reis [8], Deise Maria Furtado de Mendonça [9]

[1] Titular Professor of Clinical Anatomy, Department of Morphology, Federal University of Sergipe (UFS), Aracaju, SE, Brazil.
[2] Professor of Anatomy, Higher Education Unit of Feira de Santana (UNEF), Feira de Santana, BA, Brazil.
[3] Physiotherapy student, Higher Education Unit of Feira de Santana (UNEF), Feira de Santana, BA, Brazil.
[4] Physiotherapy Student, Centro Universitário Nobre (UNIFAN), Feira de Santana, BA, Brazil.
[5] Medical Clinic Resident of Municipal Hospital Munir Rafful (HMMR), Volta Redonda, RJ, Brazil.
[6] Medical Clinic Resident, Faculty of Medicine of São José do Rio Preto (FAMERP), São José do Rio Preto, SP, Brazil.
[7] Adjunct Professor of Human Anatomy, Department of Education and Health, Federal University of Sergipe (UFS), Sergipe, SE, Brazil.
[8] Titular Professor, Medical School of Tiradentes University (UNIT), Aracaju, SE, Brazil.
[9] Associate Professor of Human Anatomy, Department of Morphology, Federal University of Sergipe (UFS), São Cristóvão – SE, Brazil.

* Correspondence: adervalufs@gmail.com.

Citation: Aragão JA, Oliveira RF, Silva SOS, Riffel LI, Aragão ICS, Aragão FMS, Borges GR, Reis FP, Mendonça DMF. Multiple Wormian Bones in the Lambdoid Suture: A Report of Rare Occurrence. Brazilian Journal of Case Reports. 2024 Oct-Dec;04(4):141-149.

Received: 5 May 2024
Accepted: 6 June 2024
Published: 6 June 2024

Abstract: Wormian bones are irregular bony structures developed from independent ossification centers located at the junction of the sutures between the cranial bones. Although they are commonly found in healthy individuals, their presence can be associated with pathological conditions. Their number above 10, large size, or mosaic pattern are clinically considered as indicators of several pathological conditions, such as abnormalities in the central nervous system, musculoskeletal system, or metabolic changes throughout the body. Our study aims to report a rare occurrence of 40 Wormian bones found in the lambdoid suture of an adult human skull and provide a literature review. Understanding the presence of Wormian bones is crucial for professionals such as neuroanatomists, neurosurgeons, radiologists, anthropologists, and morphologists, as they may be mistaken for fractures in cases of traumatic brain injury.

Keywords: Anatomical variation; Sutural bone; Skull; Radiology; Basilar invagination.

1. Introduction

Wormian bones, also known as sutural bones, are small accessory bones found in the complex structure of the human skull. They vary in shape, number, position, and size and are often symmetrical on both sides, but can be irregular and independent. These bones were first described by Olaus Worm, a Danish doctor in the 17th century [1-3]. They are predominantly located in the cranial sutures, although they can occasionally be found in the fontanelles [4-6]. It is estimated that 8% to 15% of the global population has at least one of these bones [6-8], with a higher prevalence of location in the lambdoid suture [4, 9-11]. A special aspect is the identification of a large sutural bone in the lambda region, known as the "Inca bone". This name originated from the frequent observation of such structures in Peruvian mummies, suggesting an anthropological and historical relevance [7, 12, 13].

The presence of Wormian bones may be influenced by both genetic and environmental factors [7, 14]. There is evidence suggesting that these bones could be anatomical variations or related to pathological processes, as they are commonly observed in patients with various diseases. An important point to note is that when identified as a simple anatomical variation, Wormian bones are typically smaller in size and fewer in number. In pathological cases, it is common to identify more than 10 bones [15, 16]. For instance, an association between the presence of these bones and genetic diseases like Menkes disease, Primrose syndrome, Hajdu-Cheney syndrome, Oto-palato-digital syndrome type II, among others, has already been demonstrated [17-20]. The most well-established association is with osteogenesis imperfecta, in which the presence of Wormian bones is considered by some authors as a pathognomonic sign [21, 22]. Regarding environmental factors, a non-fusion of the cranial sutures has been suggested, as well as mechanical stresses, mainly in the occipital region [6, 22, 23].

Wormian bones are identified through imaging tests such as X-rays, computed tomography, and magnetic resonance imaging. Their presence can be noted even during the intrauterine phase through ultrasound during routine prenatal care [24]. Three-dimensional computed tomography (3D-CT) has been shown to be important for understanding the behavior of these bones for the biomechanics of the skull [6,25]. Studies with multidetector computed tomography-computed tomography angiography (MDCT-CTA) have also been reported [26].

Understanding the various causes of Wormian bones is crucial for healthcare professionals, such as clinicians, neurologists, orthopedists, and radiologists. By conducting thorough clinical and imaging assessments, it is possible to identify genetic disorders, congenital anomalies, structural abnormalities, or trauma lesions that may be associated with these bones. This knowledge is particularly important for neurosurgeons as it helps them plan surgical procedures involving the skull. Interest in Wormian bones also extends to the fields of anthropology and forensic medicine, in view of their relevance in determining identities and understanding ancient cultural practices [26-28]. Given the importance of sutural bones in both clinical and anthropological contexts, the present study aims to report the multiplicity and distribution of sutural bones in the lambdoid suture in a dry skull of an adult male and provide a literature review.

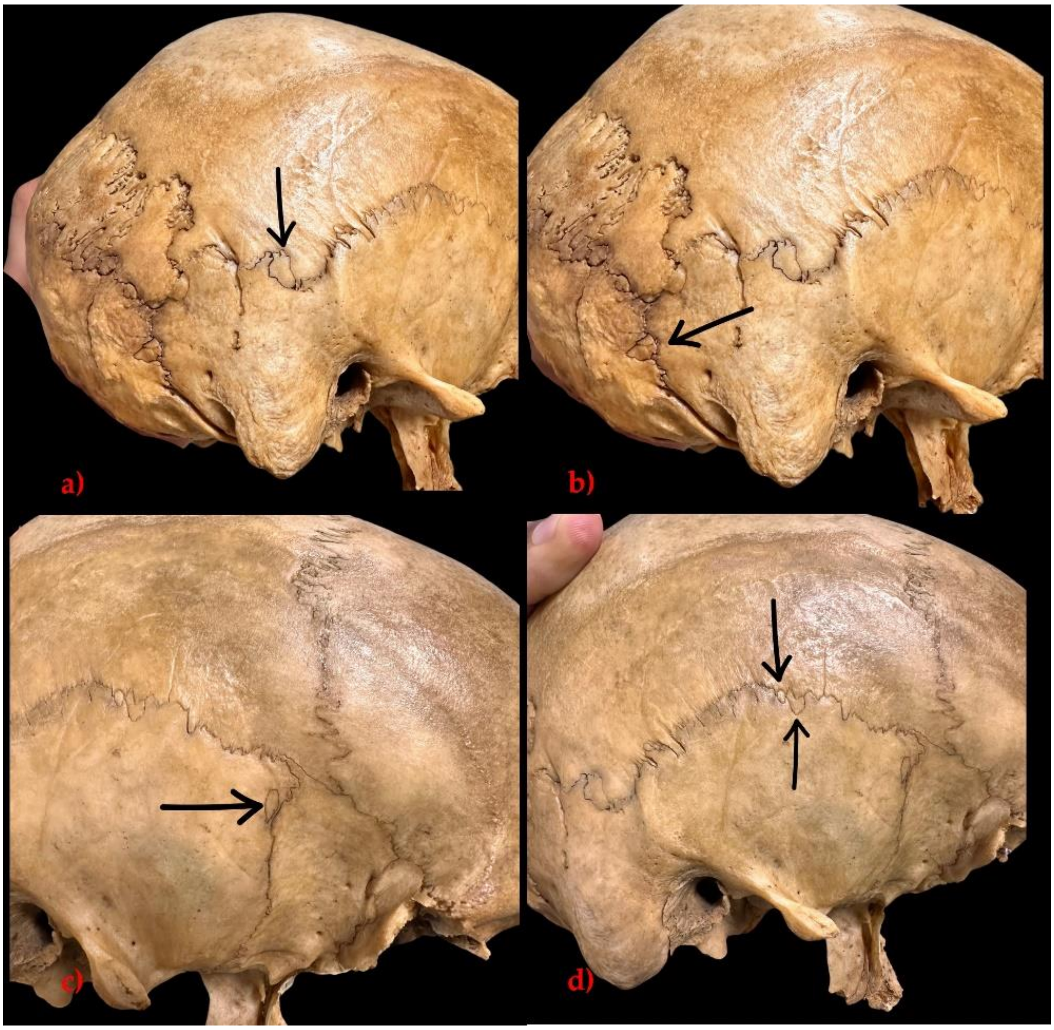
2. Case Report

During the cataloging and maintenance of specimens at the osteology museum of the Feira de Santana Higher Education Unit (UNEF), a 19-year-old male skull was identified. The skull stood out for its uniqueness due to the presentation of a notable amount of Wormian bones. The Wormian bones were located predominantly along the lambdoid suture but also in other regions of the skull. Meticulous observation was carried out to accurately determine the presence and distribution of these sutural bones. Thus, 47 Wormian bones were found, of which 40 were in the lambdoid suture (Figure 1).

Other sutures, such as the parietomastoid (Figure 2a), occipitomastoid (Figure 2b), sphenosquamous (Figure 2c), and parietosquamous (Figure 2d) contained sutural bones (Figure 2). Detailed analysis of the distribution of these bones showed notable variability, particularly, the location of these bones regarding the side of the skull. In the lambdoid suture, 19 bones were found on the right side and 21 on the left. As for the other sutures, one sutural bone was found on the right side of the following sutures: occipitomastoid, parietomastoid, and sphenosquamous; and two in the parietosquamous suture. Regarding morphology, the Wormian bones were mostly irregular in shape (40 bones), followed by triangular (4 bones) and, to a lesser extent, oval (1 bone) formats.

Figure 1. Number of Wormian bones found in the lambdoid suture.

Figure 2. Wormian bones found in other cranial sutures.



3. Discussion

Wormian bones are additional and small cranial bones that develop from separate ossification centers. They are found within sutures and fontanelles and are a very common anatomical variation. In general, the frequency of Wormian bones in the literature has ranged from 15.9% to 88.8%, with an average of 48.72%. As for location, these bones have been most frequently found at the level of the lambdoid suture with a frequency of 25% to 81.1% and an average of 60.4% (Table 1). In the present case, Wormian bones were present in 88.9% at the lambdoid suture. This finding is in accordance with the literature, which reinforces the relevance of the lambdoid suture as a point for the occurrence of these bones.

In relation to the locations of Wormian bones in other studies, the literature has shown a wide range of percentages: in the occipitomastoid suture from 3.8% to 27.14% [3,10,27,35]; in the asterion from 2.1% to 66.6% [3-5,10,29,33,35-37]; in lambda from 5.26% to 19.4% [3,5,37-39]; in the parietomastoid suture from 28.9% to 41.43% [10,35]; in the temporoparietal suture (6.1%) [27]; in bregma 1.05% (33); and in the pterion from 0.68% to 36.36% [5,12,35,37,40]. According to Aragão et al., [38], the presence of Wormian bones in the pterion region could affect access to the anterior and middle cranial fossa, particularly in surgeries for repairing middle cerebral artery aneurysms and procedures in Broca's area. In the present case, the Wormian bones, in addition to the lambdoid suture, were also located in only one bone in the sutures: parietomastoid (2.2%), occipitomastoid (2.2%), sphenosquamous (2.2%), and two in the parietosquamous suture. (4.4%). The discovery of 40 Wormian bones in the lambdoid suture in this study appears to be the highest number reported in the literature for the same suture (Table 2).

Table 1. Frequency of skulls with Wormian bones and their presence in lambdoid sutures.

Authors	Number of Skulls	Number of Skulls with Wormian bones n (%)	Number of Skulls with Wormian bonés in lambdoid sutures n (%)
Walulkar, Ksheersagar, Walulkar [29]	225	77 (34.22)	57 (74.2)
Shivaleela et al. [30]	108	47 (43.52)	36 (76.6)
Satpathi, Mangalgiri [31]	150	65 (43.33)	37 (56.9)
Showri, Suma [32]	132	74 (56.06)	60 (81.1)
Kumar, Ratna Prabha [2]	200	113 (56.5)	64 (56.63)
Natsis et al. [10]	166	124 (74.7)	74 (44.6)
Goyal, Garg, Kumar [5]	147	52 (35.3)	41 (78.84)
Nayak, Shetty [4]	27	24 (88.8)	6 (25)
Kiliç Safak, Taskin, Yücel [3]	28	12 (42.86)	7 (58.3)
Ratnaningrum [12]	69	11 (15.9)	8 (72.7)
Asharani [33]	95	24 (25.2)	13 (54.1)
Ortadeveci, Babacan [11]	29	24 (82.8)	15 (62.5)
Amadi-Ikpa et al. [13]	43	13 (30.23)	8 (61.54)
Ogut, Yildirim [34]	110	58 (52.72)	25 (43.1)
Presente estudo	1	-	1 (88.9)

Table 2. Number of Wormian bones in the lambdoid suture.

Authors	Number of Wormian bones found in the lambdoid suture
Vishali, Ebenraj, Rojomon, [39]	34
Nayak, Shetty, [4]	20
Kobayashi et al., [27]	6
Ortadeveci, Babacan, [11]	12
Present study	40

Regarding the morphology of the Wormian bones, in the present study, 88.9% were irregular in shape, 8.9% triangular, and 2.2% oval. This finding differs from Walulkar, Ksheersagar, and Walulkar [29], who reported 46.24% irregular in shape, 39.88% oval, and 13.87% triangular. Since its discovery, several hypotheses have been raised to explain the presence of Wormian bones. This has led to a large amount of information in the literature, much of it controversial. However, it appears that some points can currently be considered: (a) General development – Wormian bones have an irregular morphological appearance and are formed from independent ossification centers found along the cranial sutures and fontanelles, but the location with the highest significant frequency so far is the lambdoid suture (Table 1); (b) Prevalence – (b.1) These are very common anatomical variations (Table 1), more prevalent in early childhood; (b.2) Studies demonstrate that the younger the child age, the greater the frequency of Wormian bones [25]; (b.3) Ethnicity or cultural aspects seem to be related to the frequency of Wormian bones, as studies demonstrate an increase in occurrence in specific populations; (c) Association with diseases – The presence of more than 10 Wormian bones may be indicative of genetic or congenital disorders, and the clinician should investigate carefully when the bones are identified in the prenatal period or early childhood [9, 22, 25, 41, 42].

During development, various structures can be observed in the squamous portion of the occipital bone, which are divided into two parts: a superior and an inferior one. The first part, the interparietal segment, is a membrane bone, while the second part, the supraoccipital segment, is a cartilage bone [25, 43]. In addition to this division, three ossification centers are observed in the interparietal segment [25]. They grow similarly to other cranial bones, starting at the center and radiating outwards. This morphological organization clearly favors the formation of accessory bones at the lambdoid suture as well as the origin of additional sutures.

For Al Kaissi et al. [6], the formation of these bones results from the non-fusion of the cranial sutures, a process influenced by genetic and environmental factors. Furthermore, changes in the sutural bones, commonly identified in radiographic examinations, are attributed to the progressive softening of the sutures, like an excessively stretched dough. This process is analogously described as the stretching of pastry dough, with the lambdoid suture being particularly susceptible to this phenomenon and responsible for the development of subclinical basilar impression/invagination. The clinical relevance of changes in sutures goes beyond mere anatomical observation, with direct implications in pathological conditions such as basilar invagination.

Basilar invagination is a serious complication resulting from the invasion of the odontoid process of the axis. This invasion restricts the space in the posterior fossa of the skull, causing compression of the brain stem and spinal cord in the foramen magnum, which leads to neurological changes [6, 44]. Patients with this condition must be closely monitored to allow for timely intervention in case it progresses to neural tissue compression.

A study by Al Muroy et al. [25] evaluated CT scans from 167 patients under 2 years old with mild head trauma. The authors identified the presence of superior median fissures (21%), mendosal sutures (35%), other interparietal segment accessory sutures (9%), and interparietal sutures (6%) on the occipital bones. Wormian bones within the lambdoid suture were present in 54 children (32%). Interestingly, superior median fissures were identified in children aged 0–5 months, with 0 months being the median age. The same was observed with mendosal suture, with a median age of 1 month. Marti et al., [45] evaluated CT scans from 605 patients under 3 years old. They identified Wormian bones in 320 children (53%) and did not find a positive correlation between the presence of bones and the clinical indication that led to the imaging exam. It is worth mentioning that in this study, the maximum number of bones found per child was 8. Thus, it is possible to think that the presence of Wormian bones and additional sutures is just a part of normal development in most cases, without any pathological correlations, especially when considering the presence of less than 10 bones.

In addition to age, a factor that appears to be related to the presence of Wormian bones is ethnicity or cultural aspects, as some studies point to a high incidence in specific populations. Natsis et al., [10] evaluated 166 adult dry skulls from a Greek cohort and observed Wormian bones in 74.7% of the cases. Nayak et al. evaluated 24 adult dry skulls from a South Indian cohort and observed Wormian bones in 88.8% of the cases. However, in this last study, it must be considered that the sample was small. There are other studies that point to a high incidence in Indian populations [2, 31], as well as in Chinese [37], Nepalese [35], among other populations.

The presence of supernumerary bones has been linked to anomalies in the central nervous system, musculoskeletal system, and metabolic changes, underscoring the significance of these structures beyond their anatomical role [30,46]. Wormian bones in lambdoid suture seems to be the most significative [39]. Conditions such as Menkes disease, Primrose syndrome, Hajdu-Cheney syndrome, Oto-palato-digital syndrome type II, and particularly osteogenesis imperfecta have been identified in association with Wormian bones.

Menkes disease is a disorder caused by mutations in the ATP7A gene, which leads to reduced copper transport in the body [47]. This results in impaired function of copper-containing enzymes, affecting various tissues. Menkes disease is characterized by abnormal collagen formation, impacting blood vessels, bones, skin, hair, and the nervous system. Symptoms include sparse, kinky hair, harm to growth, hypotonia, neurological anomalies, and developmental delays [17, 47].

Primrose syndrome is a disorder caused by mutations in the ZBTB20 gene, a transcription factor that regulates neurogenesis, glucose and lipid homeostasis, and postnatal growth [48]. Symptoms include macrocephaly and a distinct facial appearance, developmental delay, with intellectual, behavioral, and physical-functional impacts, and altered glucose metabolism [18].

Oto-palato-digital syndrome type II is a genetic disorder within the Otopalatodigital spectrum disorders (OPDSD) caused by mutations in the FLNA gene. FLNA is a cytoskeletal protein that regulates cell membrane integrity and signal transduction. It interacts with integrins and second messengers, playing a role in cytoskeleton remodeling, cell shape, and migration (49). This disorder is characterized by skeletal abnormalities and, in some cases, malformations of the brain and heart [20].

Hajdu-Cheney syndrome is a disorder caused by mutations in exon 34 of the NOTCH2 gene. Notch receptors are single-pass transmembrane proteins that determine cell fate and play a critical role in skeletal development and homeostasis [50]. It is a connective tissue disease characterized by acroosteolysis of the hands and feet, developmental defects of bones, teeth, and joints, associated with progressive osteoporosis, and occasional renal abnormalities. Signs and symptoms include dolichocephaly, absence of frontal sinuses, joint laxity, dental anomalies, and short stature [19].

Osteogenesis imperfecta is a connective tissue disorder in which mutations in the *COL1A1*/*COL1A2* genes are considered one of the contributing factors to the excessive stretching typical of sutures. Null alleles, missense, or splice mutations give rise to different types of phenotypes (types I to IV) [45]. Pathogenic variants in non-collagenous genes are responsible for a smaller number of cases. These genes play various roles such as in collagen post-modification, collagen folding, intracellular trafficking, ossification, mineralization, and osteoblast development. Examples include *LEPRE1, CRTAP, PPIB, FKBP10, SERPINF1, WNT1, CREB3L1,* and *SP7* genes [51-55]. Clinically, it is characterized by bone fragility and deformity. multiple fractures, and short stature. Signals and symptoms can include brittle teeth, blue sclerae, hearing loss, impaired respiratory function, and cardiac valvular pathology.

Radiographic features of all these diseases include the presence of Wormian bones. In pathological cases, some characteristics are evident, such as the presence of ten or more bones, organization in a general mosaic-like pattern and size greater than 6 mm by 4 mm [22].

The exact process by which these bones form is not fully understood. Possibly multiple genes can be involved in their development, with their effects combining in a complex manner. The expression of these bones is likely influenced by genetic inheritance and developmental thresholds, although the specific details of these mechanisms are still being studied. Whether due to genetic causes or mechanical forces in the developing skull, pathological or induced, the deformation of the cranial joints appears to stimulate the ossification process, resulting in the presence of additional sutures, small and irregular bones near sutures, and sometimes premature closure of sutures and fontanelles.

4. Conclusion

The multiplicity of bones in the lambdoid suture found in this study illustrates the complexity and anatomical variability of Wormian bones, emphasizing the importance of these observations for significant contributions to the fields of anthropology, forensic medicine, and clinical osteology. Unfortunately, it was not possible to include further clinical data about the sample in this study, as well as any associated medical history or genetic information.

Understanding anatomical variations becomes crucial not only to expand scientific knowledge but also to rule out mistaken diagnoses of bone trauma during cranial x-rays. In this way, the present study may become relevant for professionals in areas such as radiology, orthopedics, and neurology.

Funding: This research did not receive any specific grants from public, commercial, or non-profit sector funding agencies.

Research Ethics Committee Approval: We declare that this work did not pass the ethics committee, as all cadavers found in the Human Anatomy Laboratory of the Higher Education Unit of Feira de Santana (UNEF) were obtained in accordance with Law 8,501, of November 30, 1992, which provides for the use of unclaimed corpses for the purposes of studies or scientific research and provides other measures. However, the study followed the ethical guidelines established by the Declaration of Helsinki.

Acknowledgments: The authors wish to sincerely thank those who donated their bodies to science so that anatomical research could be carried out. The results of this research can potentially improve patient care and increase the overall knowledge of humanity. Therefore, these donors and their families deserve our utmost gratitude.

Conflicts of Interest: None.

Supplementary Materials: None.

References

1. Sreekanth T, Samala N. Morphological study of wormian bones in dried adult human skulls in Telangana. Int J Anat Res. 2016 Dec;4(4):3257-62. doi: 10.16965/ijar.2016.454.
2. Kumar U, Ratna Prabha J. Wormian bones: study on dry human skulls in north Karnataka region. Int J Anat Res. 2016 Jan;4(1):1854-58. doi: 10.16965/ijar.2015.351.
3. Kiliç Safak N, Taskin RG, Yücel AH. Morphologic and morphometric evaluation of the wormian bones. Int. J. Morphol., 38(1):69-73, 2020.
4. Nayak SB, Shetty SD. High Incidence of Sutural Bones, Especially at the Asterion - A South Indian Study. Online J Health Allied Scs. 2019;18(4):8. Available at URL: https://www.ojhas.org/issue72/2019-4-8.html.
5. Goyal N, Garg A, Kumar Y. Incidence and Medicolegal Significance of Wormian Bones in Human Skulls in North India Region. Int J Appl Basic Med Res. 2019 Jul-Sep;9(3):165-168. doi: 10.4103/ijabmr.IJABMR_89_19.
6. Al Kaissi A, Ryabykh S, Ben Chehida F, Al Kaissi H, Kircher SG, Stransky MJ, Grill F. The Tomographic Study and the Phenotype of Wormian Bones. Diagnostics (Basel). 2023 Feb 24;13(5):874. doi: 10.3390/diagnostics13050874.
7. Bellary SS, Steinberg A, Mirzayan N, Shirak M, Tubbs RS, Cohen-Gadol AA, Loukas M. Wormian bones: a review. Clin Anat. 2013 Nov;26(8):922-7. doi: 10.1002/ca.22262.
8. Cirpan S, Aksu F, Mas N. The Incidence and Topographic Distribution of Sutures Including Wormian Bones in Human Skulls. J Craniofac Surg. 2015 Jul;26(5):1687-90. doi: 10.1097/SCS.0000000000001933.
9. Jeanty P, Silva SR, Turner C. Prenatal diagnosis of wormian bones. J Ultrasound Med. 2000 Dec;19(12):863-9. doi: 10.7863/jum.2000.19.12.863.

10. Natsis K, Piagkou M, Lazaridis N, Anastasopoulos N, Nousios G, Piagkos G, Loukas M. Incidence, number and topography of Wormian bones in Greek adult dry skulls. Folia Morphol (Warsz). 2019;78(2):359-370. doi: 10.5603/FM.a2018.0078.
11. Ortadeveci A, Babacan S. Prevalence, Number and Localization of Wormian Bones in Anatolian Adult Dry Skulls. Med Records. 2023;5(Suppl 1):7-10. DOI:1037990/medr.1324282.
12. Ratnaningrum SD. Identification of sutural bones in Indonesian skulls. Transl Res Anat. 2020 Mar;18:100061. doi: 1 0.1016/j.tria.2019.100061.
13. Amadi-Ikpa HA, Jacks TW, Ajie PC, Victor PD, Horsefall UA, Eyinda B, Okpara PE, Ajie PC, Reuben E, Dan-Jumbo T, Tee GP. (2023). Prevalence of Wormian Bones in Dry Adult Human Skull in University of Port Harcourt and Rivers State University. Sch Int J Anat Physiol. 2023;6(4):49-54. doi: 10.36348/sijap.2023.v06i04.003.
14. Wadanamby S, El Garwany S, Connolly D, Arundel P, Bishop NJ, DeVile CJ, Calder AD, Crowe B, Burren CP, Saraff V; Highly Specialised Service for Severe, Complex and Atypical Osteogenesis Imperfecta (NHS England); Offiah AC. Monitoring Skull Base Abnormalities in Children with Osteogenesis Imperfecta - Review of Current Practice and a Suggested Clinical Pathway. Bone. 2022 Jan;154:116235. doi: 10.1016/j.bone.2021.116235.
15. Hughes DR. Digging Up Bones. By D. R. Brothwell. London, British Museum (Natural History), 1963. xiii + 194 pp., 17 pls., 64 text-figs., 5 tables. 19s. 6d. Antiquity. 1963;37(148):327-327. doi:10.1017/S0003598X00030362.
16. Kaplan SB, Kemp SS, Oh KS. Radiographic manifestations of congenital anomalies of the skull. Radiol Clin North Am. 1991 Mar;29(2):195-218.
17. Randhawa M, Dhawan SR, Kumar S, Singanamala B, Bhatia A, Saini L, Sankhyan N. Wormian Bones and Dilated Scalp Veins in an Infant With Epilepsy. J Pediatr Neurosci. 2019 Apr-Jun;14(2):103-104. doi: 10.4103/jpn.JPN_151_18.
18. Arora V, Ferreira CR, Dua Puri R, Verma IC. Primrose Syndrome. 2021 May 6 [updated 2021 Jun 17]. In: Adam MP, Feldman J, Mirzaa GM, Pagon RA, Wallace SE, Bean LJH, Gripp KW, Amemiya A, editors. GeneReviews® [Internet]. Seattle (WA): University of Washington, Seattle; 1993–2024.
19. Zeng C, Lin Y, Lu Z, Chen Z, Jiang X, Mao X, Liu Z, Lu X, Zhang K, Yu Q, Wang X, Huang Y, Liu L. Distinct severity of phenotype in Hajdu-Cheney syndrome: a case report and literature review. BMC Musculoskelet Disord. 2020 Mar 6;21(1):154. doi: 10.1186/s12891-020-3181-0.
20. Kaissi AA, Kraschl R, Kaulfersch W, Grill F, Ganger R. Extended phenotypes in a boy and his mother with oto-palato-digital-syndrome type II. Clin Case Rep. 2015 Sep;3(9):762-6. doi: 10.1002/ccr3.317.
21. Semler O, Cheung MS, Glorieux FH, Rauch F. Wormian bones in osteogenesis imperfecta: Correlation to clinical findings and genotype. Am J Med Genet A. 2010 Jul;152A(7):1681-7. doi: 10.1002/ajmg.a.33448.
22. Cremin B, Goodman H, Spranger J, Beighton P. Wormian bones in osteogenesis imperfecta and other disorders. Skeletal Radiol. 1982;8(1):35-8. doi: 10.1007/BF00361366.
23. Sanchez-Lara PA, Graham JM Jr, Hing AV, Lee J, Cunningham M. The morphogenesis of wormian bones: a study of craniosynostosis and purposeful cranial deformation. Am J Med Genet A. 2007 Dec 15;143A(24):3243-51. doi: 10.1002/ajmg.a.32073.
24. Weissbach T, Gabis LV, Peretz-Machluf R, Reign C, Pekar Zlotin M, Spira M, Shapira M, Lev S, Elkan Miller T, Leon-Attia O, Shaham M, Weissbach A, Achiron R, Mazaki Tovi S, Weisz B, Kassif E. Clinical and neurodevelopmental outcome of prenatally diagnosed Wormian bones. Am J Obstet Gynecol. 2022 Mar;226(3):420-428. doi: 10.1016/j.ajog.2021.09.014.
25. Muroi A, Enomoto T, Ihara S, Ishikawa E, Inagaki T, Matsumura A. Developmental changes in the occipital cranial sutures of children less than 2 years of age. Childs Nerv Syst. 2021 Feb;37(2):567-572. doi: 10.1007/s00381-020-04844-8.
26. Çalışkan S, Oğuz KK, Tunalı S, Aldur MM, Erçakmak B, Sargon MF. Morphology of cranial sutures and radiologic evaluation of the variations of intersutural bones. Folia Morphol (Warsz). 2018;77(4):730-735. doi: 10.5603/FM.a2018.0030.
27. Kobayashi SBT, Soriano EP, do Nascimento EA, Campina RCF, Diniz de Carvalho MV. Frequência de ossos wormianos em esqueletos humanos brasileiros identificados. Braz J Dev. 2021 Nov;7(11): 102435-52. doi: 10.34117/bjdv7n11-050.
28. Wafae N, Ruiz CR, Pereira LA, Nunes MR, Toito E, Gomes JAJ. Análise quantitativa de ossos suturais em crânios de humanos adultos. Arq Med ABC. 2007;32(2):67-9.
29. Walulkar S, Ksheersagar D, Walulkar M. The study of wormian bones in human skulls in Vidarbha region. Panacea J Med Sci. 2012 Jul-Dec;2(2):18-21.
30. Shivaleela C, Kumar GV, Malipatil SB, Sandhya K. Morphological Study of Wormian Bones in Dried Human Skulls. Res J Pharm Biol Chem Sci. Apr-Jun 2013;4(2):584-89.
31. Satpathi DK, Mangalgiri AS. A Topographic Study of Wormian (Sutural) Bones and their Clinical Significance in the Region of Parietal Bone in Human Skulls of Central India. Indian J Forensic Med Toxicol. 2014 Jul-Dec;8(2):176-79. doi: 10.5958/0973-9130.2014.00710.5.
32. Showri R, Suma MP. Study of Wormian Bones in Adult Human Skulls. IOSR J Dent Med Sci. Dec 2016;15(12):54-60. doi: 10.9790/0853-1512055460.
33. Asharani SK. A study on the occurrence of wormian bones in adult dry skulls. Int J Anat Res. 2020 Dec;8(4.2):7814-16. doi: 10.16965/ijar.2020.228.
34. Ogut E, Yildirim FB. Wormian bone types: investigating their appearance, correlation to sex, population affinity, and clinical syndromes. Egypt J Forensic Sci. 2023 Mar;13:19. doi: 10.1186/s41935-023-00337-2.
35. Basnet LM, Shrestha S, Sapkota S. Prevalence of wormian bones in dried adult human skulls: an osteo-morphometric study in Nepal. Anat Sci Int. 2019 Jan;94(1):101-109. doi: 10.1007/s12565-018-0454-x.

36. Murlimanju BV, Prabhu LV, Ashraf CM, Kumar CG, Rai R, Maheshwari C. Morphological and topographical study of Wormian bonés in cadaver dry skulls. J Morphol Sci. 2011;28(3):176-79.
37. Li JH, Chen ZJ, Zhong WX, Yang H, Liu D, Li YK. Anatomical characteristics and significance of the metopism and Wormian bones in dry adult-Chinese skulls. Folia Morphol (Warsz). 2023;82(1):166-175. doi: 10.5603/FM.a2022.0006.
38. Aragão JA, de Oliveira Carvalho C, Barreto PG, Sant'Anna Aragão FM, Sant'Anna Aragão IC, de Godoy JRP, Reis FP. Epipteric Bones in the Pterion in Dry Human Crania from a Region of Northeastern Brazil: Morphological and Morphometric Study. International Journal of Anatomy, Radiology and Surgery. 2016 Oct; 5(4):AO08-AO11. doi: 10.7860/IJARS/2016/21959:2195.
39. Vishali N, Ebenraj TJ, Rojomon TC. A rare existence of significant number of Wormian bones in the lambdoid suture. Int J Sci Res. 2014 Aug;3(8):671-77.
40. Uchewa OO, Egwu OA, Egwu AJ, Nwajagu GI. Incidence of wormian bones in the dried skull of Nigerian males. Int J Anat Var. 2018 Mar;11(1):32-4.
41. Pryles CV, Khan AJ. Wormian bones. A marker of CNS abnormality? Am J Dis Child. 1979 Apr;133(4):380-2.
42. Kozerska M, Skrzat J, Walocha J, Wrobel A, Leszczynski B. Imaging of the Wormian bones using microcomputed tomography. Folia Med Cracov. 2013;53(4):21-8.
43. Srivastava HC (1992) Ossification of the membranous portion of the squamous part of the occipital bone in man. J Anat 180(Pt 2): 219–224. https://doi.org/10.1111/(ISSN)1469-7580
44. De Nova-García MJ, Sola RG, Burgueño-Torres L. Influence of the Severity of Osteogenesis Imperfecta on Cranial Measurements. Children (Basel). 2023 Jun 8;10(6):1029. doi: 10.3390/children10061029.
45. Marti B, Sirinelli D, Maurin L, Carpentier E. Wormian bones in a general paediatric population. Diagn Interv Imaging. 2013 Apr;94(4):428-32. doi: 10.1016/j.diii.2013.01.001.
46. Singh R. Wormian Bones: Prevalence, Topography, and Implications. J Craniofac Surg. 2024 Jan-Feb 01;35(1):247-250. doi: 10.1097/SCS.0000000000009746.
47. Asharani SK. A study on the occurrence of wormian bones in adult dry skulls. Int J Anat Res. 2020 Dec;8(4.2):7814-16. doi: 10.16965/ijar.2020.228.
48. Arora V, Leon E, Diaz J, Hove HB, Carvalho DR, Kurosawa K, Nishimura N, Nishimura G, Saxena R, Ferreira C, Puri RD, Verma IC. Unique skeletal manifestations in patients with Primrose syndrome. Eur J Med Genet. 2020 Aug;63(8):103967. doi: 10.1016/j.ejmg.2020.103967.
49. Zaytoun GM, Harboyan G, Kabalan W. The oto-palato-digital syndrome: variable clinical expressions. Otolaryngol Head Neck Surg. 2002 Feb;126(2):129-40. doi: 10.1067/mhn.2002.122184.
50. Canalis E, Zanotti S. Hajdu-Cheney syndrome: a review. Orphanet J Rare Dis. 2014 Dec 10;9:200. doi: 10.1186/s13023-014-0200-y.
51. Forlino A., Marini J.C. Osteogenesis imperfecta. Lancet. 2016;387:1657–1671. doi: 10.1016/S0140-6736(15)00728-X.
52. Palomo T, Vilaça T, Lazaretti-Castro M. Osteogenesis imperfecta: diagnosis and treatment. Curr Opin Endocrinol Diabetes Obes. 2017 Dec;24(6):381-388. doi: 10.1097/MED.0000000000000367.
53. Marom R, Rabenhorst BM, Morello R. Osteogenesis imperfecta: an update on clinical features and therapies. Eur J Endocrinol. 2020 Oct;183(4):R95-R106. doi: 10.1530/EJE-20-0299.
54. Deguchi M, Tsuji S, Katsura D, Kasahara K, Kimura F, Murakami T. Current Overview of Osteogenesis Imperfecta. Medicina (Kaunas). 2021 May 10;57(5):464. doi: 10.3390/medicina57050464.
55. Rossi V, Lee B, Marom R. Osteogenesis imperfecta: advancements in genetics and treatment. Curr Opin Pediatr. 2019 Dec;31(6):708-715. doi: 10.1097/MOP.0000000000000813.