Morphological analysis of intrasutural bones in human skulls from laboratories in Paraíba state (Brazil)

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***Abstract*—Introduction: The neurocranium is mostly arranged by fibrous intertwined sutures, in which sesamoid bones called wormian or intrasutural bones (ISB) may appear. Wormian bones prevalence in the population varies all over the world, and there are few reports about its frequency in the Brazilian population. To analyze frequency, variations, morphometry, and sexual dimorphism of ISB in human skulls from university laboratories in the state of Paraíba (Brazil).**

Material and Methods: 115 dry skulls from three anatomy laboratories at universities in Paraíba were used for the study. ISB presence and location were analyzed by photographic assessment. The maximum vertical (MVD) and maximum horizontal (MHD) distances were obtained using a carbon fiber digital caliper (Nove54). To measure the vertical curved distance (VCD) and horizontal curve (HCD), a small fabric thread was used, and it was compared to a millimeter ruler for measurement.

Results: Of the 63 evaluated skulls, 40 (63.49%) had at least one ISB. There was a predominance of females (34.9%), the most common shape was irregular (42.38%) and the most frequent location was the lambdoid suture (48.9%). MVD ranged from 2.7 to 25.5 mm, and MHD between 2.8 and 44.5 mm. HCD ranged between 1 mm and 48 mm, and VCD between 1 and 25 mm. Sexual dimorphism did not influence morphometric characteristics.

Conclusion: Most skulls studied had intrasutural bones, with slight predominance in female skulls, but without sexual dimorphism. The most frequent site was in the lambdoid suture.

Keywords—cranial sutures, skull, wormian bones.

I. INTRODUCTION

The skull corresponds to the bone structure of the head, responsible for supporting and protecting sensory organs and part of the central nervous system. During the ossification process, it is subdivided into the viscerocranium, which comprises the facial bones and, during the embryonic phase, it originates from the mesenchyme of the first three pharyngeal arches; and the neurocranium that surrounds the brain and develops from the surrounding mesenchyme [1]. The neurocranium is composed by bones joined by fibrous intertwined sutures, whose development arises from intramembranous ossification and endochondral ossification, subdivided into the chondrocranium, which corresponds to the base of the skull, and calvaria or cranial vault, which constitutes the skull roof [2].

Among the sutures, it is possible to find the intrasutural bones (ISB), also called wormian bones (WB) or sutural bones. Its name is credited to the Danish physician Olaus Wormius, in the beginning of the 17th century, although the description has already been made before, by Paracelsus (1460-1541), who named a bone in the posterior fontanelle as “ossiculum antiepilepticum”, due to a supposed relationship with epilepsy [3,4].

Wormian bones are sesamoid, asymmetrical, irregular structures, with variable morphology and thickness, located mainly between the sutures, generally between those where a fontanelle was previously located [5]. Its formation comes from isolated ossification centers along the skull, because of several factors, such as genetic predisposition, external mechanical pressure or craniosynostosis [6].

The prevalence of ISB in the population ranges from 8 to 15%, it is more frequent in the Chinese population and in males [7–10]. In a survey carried out with dry skulls from different populations around the world, a percentage of 80% of ISB was observed among the Chinese and 55% in the Anglo-Saxon population [11].

The presence of ISB has been associated with some types of cranial malformations, endocrine syndromes, and genetic syndromes. Thus, the mnemonic PORKCHOPS was created, which corresponds to the changes associated with a higher frequency of ISB: pycnodysostosis, trichopolyodystrophy, cleidocranial dysostosis, rickets, congenital hypothyroidism, otopalatodigital syndrome, primary acral osteolysis, Down syndrome and osteogenesis imperfecta [10,12,13].

There are few reports about the frequency of ISB in the Brazilian population. Furthermore, in view of the country's intense miscegenation, it is important to consider existing differences between the different regions of the country. No study evaluating the population of Paraíba has been documented in the literature.

Considering the relevance of morphometric studies, this study aimed to analyze the frequency, variations, morphometry and sexual dimorphism of ISB in human skulls from university laboratories in the state of Paraíba (Brazil).

II. MATERIAL AND METHODS

This is a cross-sectional study, with primary data collection. The sample consisted of 115 dry skulls, from a survey carried out in the anatomy laboratories of three universities in Paraíba. For this study, intact skulls, that presented the third molar or vestige, of both sexes were considered. Bones damaged by accident, skulls with obliterated sutures and children's skulls were excluded from the sample. Data collection was carried out from April to June 2021.

The presence and location of ISB were verified through direct evaluation and recorded by photographs, taken with a Nikon Coolpix P600 digital camera. Initially, the criteria proposed by Vanrell (2002) [14] were applied to assess sexual dimorphism in dry skulls. These include evaluation of the forehead, glabella, frontonasal junction, supra-orbital margins, mastoid apophyses, weight, occipital condyles, and mastoid and styloid processes [14]. Subsequently, information on the number of ISB was documented, each one characterized according to its topography and format.

Information on ISB dimensions was also obtained, measured in vertical and horizontal extensions. The maximum vertical (MVD) and maximum horizontal (MHD) distances were obtained using a carbon fiber digital caliper (Nove54). To measure the vertical curve distance (VCD) and horizontal curve distance (HCD), a fabric thread segment was used, which was then compared to a millimeter ruler.

Data were submitted to double entry, whose database validation was verified using the “validate” tool, using the statistical program Epi Info 6.04 (Centers for Disease Control and Prevention, Atlanta, United States). Results were grouped according to sex. Numeric data are presented as mean and standard deviation, minimum and maximum values, and categorical data through frequency distribution. Data normality distribution was verified by the Kolmogorov-Smirnov test. To compare the morphometric characteristics of ISB between males and females, the Mann-Whitney test was used. In all analyses, a confidence interval of 95% (95% CI) and statistical significance of $p<0.05$ were considered..

III. RESULTS

Of the 115 dry skulls evaluated, 92 presented ISB (%) and 23 had more than one ISB, ranging from 2 to 8 bones in the same skull, resulting in 92 bones included for analysis. Table 1 presents the characteristics regarding presence, location, and shape of ISB in both sexes.

ISB frequency was slightly higher in female skulls (34.9%) compared to males (28.57%). Regarding the location of the bones, there was a higher frequency in the regions of the lambdoid suture (48.9%) and the asterion (20.64%) (figure 1), in both sexes, followed by the parietomastoid suture (8.6 %), lambda (5.4%) and sphenozygomatic suture (5.38%). In female skulls, there was a higher frequency of ISB in the occipitomastoid and sphenozygomatic sutures compared to men. The ISB shape ranged from irregular (42.38%), quadrangular (40.21%) and triangular (17.4%). Irregular ones were more frequent in males (26.08%), whereas quadrangular ones were more common in females (21.74%). Triangular bones had a similar distribution for both sexes (8.7%).

Table 1: Dimorphic morphological features of intrastural bones of dry human skulls.

Variable	Male		Female		Total
	N	F (%)	N	F (%)	Fac (%)
ISB presence					
Yes	50	43.4	42	36.5	79.9
No	11	9.5	12	10.4	19.9
Total	61	53.04	54	46.96	100
Skull shapes with ISB					
Rounded	36	39.13	29	31.52	70.65
Pentagonal	14	15.21	13	14.13	29.34
Total	50	54.34	42	45.65	100
ISB Location					
Lambdoid	24	26.08	21	22.82	48.9
Asterion	12	13.04	7	7.6	20.64
Parietomastoid	4	4.3	4	4.3	8.6
Frontozygomatic	1	1.08	0	0	1.08
Occipitomastoid	1	1.08	3	3.26	4.34
Coronal	2	2.17	0	0	2.17
Lambda	3	3.26	2	2.17	5.43
Pterion	1	1.08	2	2.17	3.25
Sphenozygomatic	1	1.08	4	4.3	5.38
Total	49	53.26	43	46.73	100
ISB shape					
Triangular	8	8.7	8	8.7	17.4
Quadrangular	17	18.47	20	21.74	40.21
Irregular	24	26.08	15	16.30	42.38
Total	49	52.17	43	47.82	100

Characteristics of studies that assessed the presence of persistent median artery and its possible relationship with carpal tunnel syndrome.

Figure 1: Wormian bones in lambdoid suture (left) and asterion (right).

Table 2 presents the morphometric characteristics of the intrasutural bones, according to sexual dimorphism. MVD in ISB ranged from 2.7 to 25.5 mm and MHD between 2.8 and 44.5 mm. HCD presented a minimum distance of 1 mm and a maximum distance of 48 mm and the VCD of 1 and 25 mm, respectively. Although slightly higher values were observed in female skulls, no significant difference was observed compared to males.

Table 2: Dimorphic morphometric features in intrasutural bones of dry human skulls.

Variable	Male		Female		
	Vmin - Vmax	Mean ± SD	Vmin - Vmax	Mean ± SD	*p*
MVD	2.7 – 25.5	12.09 ± 5.9	1.7 – 47.6	12.8 ± 10.5	0.64 (ns)
MHD	2.8 – 44.5	12.93 ± 7	2 – 53.3	13.29 ± 11	0.34 (ns)
VCD	1 - 25	13.3 ± 6.1	2 - 55	15.68 ± 11	0.90 (ns)
HCD	1 - 48	13.5 ± 8.3	1 - 64	16.2 ± 2.4	0.71 (ns)

MVD – maximum vertical distance; MHD – maximum horizontal distance; VCD – vertical curve distance; HCD – horizontal curve distance; ns – non-significant for Mann-Whitney test.

Table 3 summarize data from the studies that documented the presence of ISB in dry skulls.

Table 03: Comparison of the intrasutural bones frequency documented in studies with dry human skulls.

Authors (year)	Study setting	n	FAC (%) TOTAL	Most frequent location
Brito et al. (2021)	Paraíba (Brazil)	115	80.00	Lambdoid
Henríquez-Pino et al. (1992) [8]	Brazil (not specified)	200	44.00	Lambdoid
Wafae et al. (2007) [9]	São Paulo (Brazil)	272	36.00	"between parietal and occipital bones"
Murlimanju et al. (2011) [3]	Mangalore (India)	78	73.10	Lambdoid
Patil & Sheelavant (2012) [20]	Banglalore (India)	180	52.22	Lambdoid
De Almeida et al. (2012) [21]	São Paulo (Brazil)	35	40.00	Lambdoid
Walulkar et al. (2012) [22]	Maharashtra (India)	225	34.22	Lambdoid
Shivaleela et al. (2013) [23]	Karnataka (India)	108	43.53	Lambdoid
Sadhu et al. (2013) [24]	West Bengal (India)	111	69.37	Lambdoid
Patel et al. (2015) [25]	Gujarat (India)	27	44.40	Lambdoid
Cirpan et al. (2015) [26]	Izmir (Turkey)	150	59.30	Lambdoid

Halagatti & Channabasanagouda (2016) [15]	India (South)	110	69.09	Lambdoid
V.G. et al. (2017) [16]	Karnataka (India)	200	61.50	Lambdoid
Shantharam & Manjunath (2017) [17]	Not specified	110	81.89	Lambdoid
Baa et al. (2018) [31]	Andhra Pradesh (India)	110	70.80	Asterion
Uchewa et al. (2018) [18]	Ebonyi (Nigeria)	22	45.46	Lambdoid
Natsis et al. (2019) [6]	Zografou and Macedon Central (Greece)	166	74.70	Lambdoid
Basnet et al. (2019) [19]	Pokhara (Nepal)	70	88.57	Lambdoid

IV. DISCUSSION

Results reported in this study show high prevalence of ISB in human dry skulls. Different factors seem to have implications for the formation of ISB. O'Loughlin (2004) suggests that the formation of these bones in anterior positions is likely related to genetic factors, and the posterior ones seem to be linked to environmental factors [15]. There are implications for ISB formation according to the degree of deformity in the skull. In skulls with asymmetric deformation, a greater number of ISBs was observed on the side of greater deformation [16].

There is a predominant presence of ISB in the lambdoid suture, which seems to be related to brain expansion [3,6,8,17–28]. Cranial deformations in a single direction, especially in the anteroposterior direction, lead to greater ISB formation compared to those that occur uniformly and circumferentially, when the compression forces are evenly distributed, which generates insufficient conditions for ISB development [16]. As the posterior part of the brain grows faster compared to the anterior portion, this generates greater stress and distance forces between the parietal and occipital bones, so that their separation facilitates ISB formation between them [15]. Thus, intracranial disorders such as hydrocephalus, which result in expansion of the sutures, especially in the early years, where the bones are more malleable, would result in the formation of ISB [29]. The action of external forces can also play a role in this process, considering that, during development, the contraction of the temporal and trapezius muscles pulls the occipital and parietal bones, pushing them apart [30]. Therefore, it is possible that the number of ISB is associated with severe cranial deformities, especially in the occipital region [29].

Studies have observed an association of ISB with osteogenesis imperfecta [16,32–35], so that the presence of these structures on radiology can be useful for the diagnosis of this condition [34]. Cremin et al. (1982) consider the presence of at least 10 ISB as indicative of possible osteogenesis imperfecta [35]. Osteogenesis imperfecta is a genetic disorder that results in changes in type I collagen [34]. Its association with ISB formation is the result of the gradual deformity of bones of the skull base which, due to the deficiency of type I collagen, become softer, so that it cannot support the weight of the brain structures, leading to a condition called basilar abnormality. This was most frequently observed in type III and type IV osteogenesis imperfecta [36]. Although the occurrence of ISB is not specific to this condition [16], the observation of these structures in younger children may be indicative of some developmental abnormality, and further investigation is important, particularly regarding bone dysplasia. Furthermore, in the field of forensic medicine, knowledge of these cranial variations is important to clarify unexplained fractures when there is doubt whether they are being caused by osteogenesis imperfecta or by physical abuse [10].

A relevant feature about ISB anatomy and morphometry is the interpretation of radiological findings. ISB presence may lead to confusion in the evaluation of images, especially in posterior head injuries with suspected multiple fractures [37] and in the pterium region, where ISB are known as "pterium ossicle", "epiteric bone" or "bone of Flower" [38]. The presence of variations in the asterium also plays a role in the planning and guidance of surgeries in the posterior cranial fossa [39].

In this study, there was higher frequency of ISB in females compared to males, and it was more frequent in lambdoid suture, in both genders, but there was no statistically relevant sexual dimorphism. These findings were also observed in other studies [6,9,22,26]. On the other hand, two studies in India found higher prevalence of ISB in males.

The results obtained for MHD (12.93±7 mm for men and 13.29±11 mm for women) and MVD (12.09±5.9 for men and 12.08±10.5) are equivalent to the study of DAS S etc. (2012), who found 11 mm in MHD and 15 mm in MVD. Our results are also accordingly with the definitions that characterize ISB sizes as ranging from less than 1 mm to 50-90 mm [40].

ISB measurements were performed in case studies. Atlanoğlu et al. (2016) found 6.5 x 6 cm and 4 x 4 cm bones in the anterior fontanelle in a computed tomography [41], while Samson et al. (2008) reported a 7 x 8 cm bone at the same location [42]. Both studies did not analyze sexual dimorphism.

Regarding the bones' shape, our results showed that the most frequent shape was irregular (42.38%), of which more than half were male (26.08%), whereas females predominantly presented quadrangular shape (21.74%). The general results corroborate other studies, such as the one by Reddy (2018), which documented the irregular shape in 16.66% [43]; Vasanthi et al. (2015), whose detection of irregular ISB was 100% [44], and De Heus G (2018), that reported irregular ISB in 43.16% in children's skulls analyzed using computed tomography [45]. On the other hand, results of Showri & Suma (2016) are conflicting, because the quadrangular shape was the most frequent format, in 52.70% of individuals [5].

V. CONCLUSION

Most investigated skulls presented ISB, with a slight predominance in female skulls. However, there was no sexual dimorphism regarding morphometric aspects. The most affected site was the lambdoid suture, corroborating findings from previous studies.

Intrasutural bones have clinical-surgical, anthropological, and medico-legal relevance. Most studies have been carried out in the East (particularly India), and current data on Brazil and nearby regions are still insufficient. This study reaches the objective of contributing to the global statistics about these structures, however, other local studies are still needed to better clarify the morphological data on ISB..

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