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ANATOMIC VARIATIONS OF CALVARIAL INTRASUTURAL BONES: AN AUTOPSY STUDY

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ABSTRACT

Calvarial intrasutural bones (CIB) incidences are known to vary in different populations. The objective of this study was to determine the frequency, location, number and gender difference if any, of CIB in Nigerian skulls. This descriptive observational study included 96 adult skulls. Out of 96 subjects, 58 were males and 38 were females (M:F=1.5:1). The mean age was 50.2±16.5 years (19-83 years). Most of the skulls studied (56%) had CIB. Sixty percent of the males and 50% of the females had CIB (p=0.318). The lambdoid suture (33.3%) and lambda (20.8%) area were the most common sites for CIB. When present, CIB were mainly on the right (43.8%). The bregma, pterion and coronal suture were rare sites for CIB in this study population. These results established that CIB are common in adult Nigerian skulls and that there is no significant association between presence of CIB with gender or age. This variation should be taken into consideration in patient evaluation and surgical planning.

Key words: *Asterion, Calvarial intrasutural bones, Lambda, Lambdoid suture, Sagittal suture*

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INTRODUCTION

Calvarial intrasutural bones (CIB) are supernumerary or accessory bones found within cranial sutures or fontanelles (Bellary et al., 2013; Cipran et al., 2015; Khan et al., 2011). These bones are generally said to be normal variants. Though CIB have no regular relation to normal ossification centres, their development has been described as resulting from isolated island of intramembranous ossifications in the neurocranium as independent ossification centres in the calvaria in addition to the normal ossification centres (Bellary et al., 2013; Cipran et al., 2015; Jeanty et al., 2000). The mechanism involved in the formation of sutural bones has been suggested to result from artificial deformation of skulls. Some authors' have hypothesized that its formation is regulated by genetic factors (Wilczak and Ousley, 2009; Murphy, 1956). These accessory bones are associated with insufficient rate of suture closure and regarded as epigenetic and hypostatic genes.

The location, number, shape and size of CIB also differ from skull to skull. There is population and gender variation with respect to incidence (Europeans, Asians and Americans); incidences of 10-80% in different populations have been reported (Jeanty et al., 2000; Marti et al., 2013). Calvarial intrasutural

bones are stated to be more common in males than females (Pedro et al., 2007; Bryan et al., 1982; Nayak et al., 2008). Previous studies have noted large number of CIB at the lambda and along the lambdoid suture (Seema and Mahajan, 2014). According to Bergman et al. (1998), the most common site for CIB is the lambdoid suture area, with about 40% of skulls having CIB (Bergman et al., 1988).

The importance of CIB is evident in anthropology, developmental and comparative anatomy, neuroradiology, neurological surgery and forensic medicine. Calvarial intrasutural bones have been used in the assessment of the differences between population groups based on peculiarities in different populations. Knowledge of the presence of CIB is of radiological importance as they may be mistaken for skull fracture during radiological assessment of the head in calvarial trauma (Khan et al., 2011; Sexena et al., 1988). Marti et al noted that when CIB exceed 10 in an individual it may indicate the presence of an underlying craniocerebral pathology (Marti et al., 2013). However, there have been cases where there were no craniocerebral diseases associated with the CIB (Wilczak and Ousley, 2009). The presence of CIB bones may lead to complications during fashioning of burr holes or calvarial instrumentation (Keskil, 2003; Kumar et al., 2016; Ersoy et al., 2003) in neurosurgical procedures. The variations of these bones can be further evidence in the identification of a deceased person when they are compared with available ante mortem records.

The aim of the study is to determine frequency, location and number of CIB in Nigerian skulls and to analyse them topographically.

MATERIALS AND METHODS

The study was in accordance with the ethical standards of the Ethical Committee of our Institutions and in accordance with the 1964 Helsinki declaration and its later amendments. The approval to perform this study was obtained from tour institutional Health Research and Ethics Committee (LREC/06/10/1232).

The target population for this study were Nigerian adults 18 years and above who had consecutive post-mortem examination at the Pathology and Forensic Medicine, Lagos State University Teaching Hospital, Ikeja, Lagos State, Nigeria. A total of 96 skulls were examined. All patients that were less than 18 years and non- Nigerians were excluded.

In addition, skulls with skull fractures and those with previous cranial surgeries were excluded. Information regarding demography and medical history were obtained from the case notes. All skulls were exposed by subperiosteal dissection so as to adequately visualize the suture areas. The frequency, number and topographic distribution of CIB were then documented. The length and breadth of the intrasutural bones were also measured.

The collated data were statistically analysed using percentage in each category for nominal variables (gender, location of CIB, and side of CIB). Means, standard deviations and range were obtained as appropriate. Unpaired Student's t-tests were used to determine any differences between males and females means. Correlation of CIB presence with gender and age was investigated using the Chi square test (or Fisher's exact test) while side asymmetry was examined with McNemar test. The level of significance was fixed at less than 5% probability for chance. Data was analysed using Statistical Package for the Social Sciences (SPSS) computer software Version 23 IBM.

RESULTS

Out of 96 subjects studied, 58 were males and 38 were females (M: F=1.5:1). The mean age was 50±16.5 years (19-83 years) (Table 1). A least 1 CIB was present in 54 subjects (56.3%). Calvarial intrasutural bones were noted in 35 males (60.3%) and 19 females (50%) (p= 0.318). Thirty-two individuals (33.3%) had lambdoid suture sutural bone(s) while 20 (20.8%) and 10 (10.4%) had sutural bones at the lambda and asterion respectively (Table 2). There was no significant difference between CIB presence and age group (p= 0.920) or gender (p= 0.318) (Table 2). Neither was there significant association between the presence of intrasutural bones in the lambdoid suture and lambda (p= 0.415), asterion (p= 0.512) or squamous suture (p= 0.227). There was also no significant association between CIB presence in the lambda and sagittal suture (p= 0.594).

A total of 132 sutural bones were found; 63 found on the right (42 subjects), 43 found on the left (23 subjects) and 26 found in the midline (23 subjects). Lambdoid suture had the highest number of intrasutural bones i.e. 80 in 32 subjects (33.3%). This is followed in order of frequency by the lambda with 22 in 20 subjects (20.8%), asterion with 16 in 10 subjects (10.4%), squamous suture with 10 in 6 subjects (6.3%) and sagittal suture with 4 in 3 subjects (3.1%). There was no intrasutural bone found in pterion, bregma and coronal suture.

The right side was the most common site (42 individuals, 43.8%) for CIB. On the left side and midline, 26 individuals (27.1%) each had CIB. Of the sutural bones occurring on the right, the lambdoid, asterion and squamous suture accounted for 31.25%, 9.38% and 4.12% respectively. On the left side, CIB are present in the lambdoid, asterion and squamous suture of individuals in 17.71%, 5.21% and 6.25% respectively. In the midline, most of the 26 CIB were present in the lambdoid (20 patients, 20.8%).

Two autopsy subjects (2.1%) had 10 or more intrasutural bones. The largest sutural bone was 12 X 7cm (Table 3). This was found in the lambda (Inca bone). There was no statistically significant association between presence of CIB and side.

Table 1: Socio-demographic characteristics

Frequency	**Frequency (=96)**	**Percentage**
Gender		
Male	58	60.4
Female	38	39.4
Age group (years)		
≤45	44	45.8
46-65	32	33.3
≥66	20	20.8
Mean± SD	50.19±16.5	

Table 2: Distribution of Intrasutural Bones

S/N	**Location**	**No of Subjects with Intrasutural Bones (%)**	**No of Intra-sutural Bones**	**Right**	**Left**
1	Sagittal suture	3 (3.1)	4	NA	NA
2	Lambda	20 (20.8)	22	NA	NA
3	Bregma	0 (0.0)	0	NA	NA
4	Lambdoid Suture	32 (33.3)	80	48	32
5	Squamous Suture	6 (6.3)	10	4	6
6	Asterion	10 (10.4)	16	11	5
7	Pterion	0 (0.0)	0	0	0
8	Coronal Suture	0 (0.0)	0	0	0
TOTAL		54	144	63	43

*NA- Not applicable

Table 3: Length and Breadth of Calvarial Intrasutural Bones

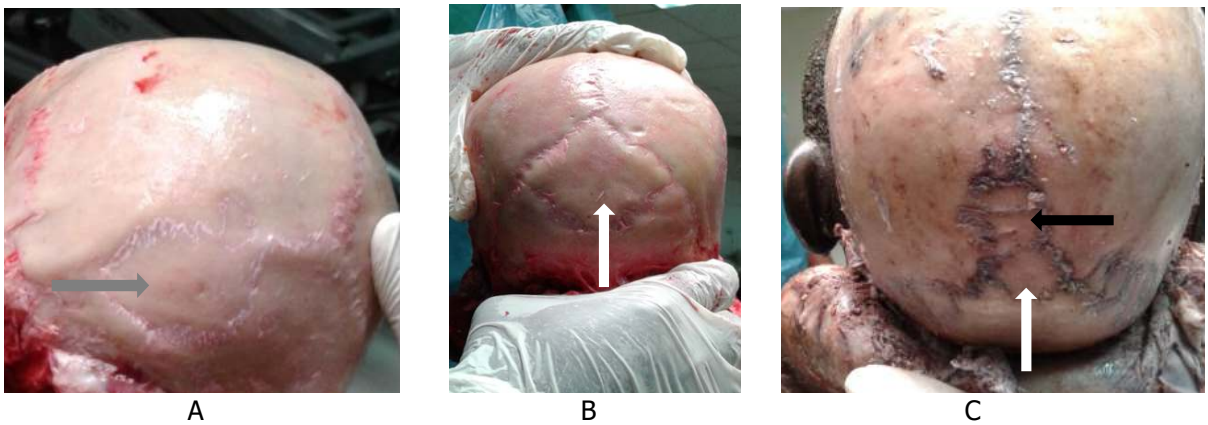
Location	**Median Length in cm**	**Median Breadth in cm**
Sagittal Suture	2.8 (1-3.5)	1.0 (1.0-2.5)
Squamous Suture	3.5 (2-6.5)	2.0 (1.0-2.5)
Lambdoid Suture	1.5 (0.4-7.0)	1.0 (0.2-5.0)
Lambda	3.0 (0.8-12.0)	2.5 (1.4-7.0)
Asterion	1.5(0.5-2.0)	1.0 (0.5-2.0)

Table 4: Association between Intrasutural Bones presence and Demographic Characteristics

Variable	Present	Absent	p-value
Gender			0.318
Male	35 (60.3%)	23 (39.7%)	
Female	19 (50.0%)	19 (50.0%)	
Age Group (years)			0.920
<45	24 (54.5%)	20 (45.5%)	
45-65	18 (56.2%)	14 (43.8%)	
≥66	12 (60.0%)	8 (40.0%)	

Table 5: Relationship of gender to Calvarial location of Intrasutural bones

Variable	Male	Female	p-value
Squamous suture			
None	54(93.1)	36(94.7)	
Present	4(6.9)	2(5.3)	0.746
Lamboid suture			
None	36(62.1)	28(73.7)	
Present	22(37.9)	10(26.3)	0.238
Asterion			
None	50(86.2)	36(94.7)	
Present	8(13.8)	2(5.3)	0.181



A B C

Figure 1 A, B, C: Calvarial intrasutural bones in lambdoid suture (gray arrow), Lambda (white arrow) and posterior sagittal suture (black arrow).

DISCUSSION

The human skull develops from neurocranium and viscerocranium with the calvaria formed primarily in the neurocranium. Calvarial intrasutural bones were first described by Paracelsus (1460 to 1541 CE (Jeanty et al., 2000) while the specific narrative of lambdoid sutural bones also called, Wormian bones was by a Danish Professor of anatomy, Olaus Worm (1588-1654). Olaus Worm designated these bones in a letter to Thomas Bartholin, and Bartholin named them Ossa Wormiana, their present name (Siddiqui, 2013).

Calvarial intrasutural bones vary from about 8-15% in some populations to about 55% in Anglo-Saxon population, 75% amongst Greeks and 80% in the Chinese (Pedro et al., 2007; Natsis et al., 2019; Marti et al., 2013). In Indian and Nigerian populations, incidences of 11.79% and 3.79% respectively have been reported (Sexena et al., 1986). In this Nigerian study wherein, we could definitely limit the samples to Nigerians, we found 56.3% incidence. This is far different from the previous osteology Nigerian study but similar to Kumar and his colleague reported incidence of 56.5% of CIB in 200 skulls (Kumar et al., 2016). Acquaintances of CIB in more than 50% of the skulls studied in our population is to be kept in view by Radiologists during calvarial radiological assessment, Neurosurgeons when making burr holes for various intracranial and extracranial procedures, and by forensic Anthropologists when trying to identify a person's race using the skull.

Philippe Jeanty et al and Sanchez-Lara et al separately reported intrasutural bones as being more common on the right (Jeanty et al., 2000; Sanchez-Lara et al., 2007) while Cirpan et al reported higher intrasutural bones

on the left (Cirpan et al., 2015). In our study, CIB were found mostly on the right 63(47.7%). On the left and the midline they were found in 43(32.6%) and 26(19.7%) respectively. Side difference was however not statistically significant, neither was gender and age difference affected the incidence of CIB (Table 4, 5). This is similar to Natsis et al study (Natsis et al., 2019).

Most CIB were located within the lambdoid suture (64 skulls, 56.63%) (Kumar et al., 2016). This is similar to our study, though we found a relatively lower frequency of 33.3% (32 patients). Calvarial intrasutural bones were most commonly located in the lambdoid suture (44.6%), followed in order of frequency by the coronal suture (39.8%), asterion (21% on the left and 15.3% on the right side) and parietomastoid suture (15.1% on the left and 13.9% on the right side) in Natsis et al study (Natsis et al., 2019). He noted CIB bones within the occipitomastoid, sagittal, squamosal, zygomaticosphenoid, metopic, frontonasal and frontozygomatic sutures.

Intrasutural bones can also be present at the region of the lambda. These can be classified as interparietal or pre interparietal sutural bones. Interparietal sutural bones have their lower edges situated slightly above the external occipital protuberance. These bones can be single or multiple. It is sometimes called the inca bone (Sexena et al., 1986). In a study conducted on 40 adult Nigerian skulls, 20% were noted to have CIB in the lambda region (Sexena et al., 1986). In our study we found 21% of patients with CIB in this region.

When a CIB is present at the pterion (junction of the parietal, frontal, greater wing of the sphenoid, and squamous portion of the temporal bones) it is called Pterion ossicles, epipteric bone or Flower's bone (Malhata et al., 1978; Das et al., 2009). This was the next most common site with an incidence of 11.79% of epipteric bone in Indians (Jeanty et al., 2000). Epipteric bone was noted in 3.6% (Ukoha et al., 2013) and 0% (Adejuwon et al., 2013) of dried skulls in a previous study from Nigeria. Four percent (left side only) was noted in a study from Turkey (Oguz et al., 2004) and 10% from Japan (Matsumura et al., 1991). None of the skulls we examined had epipteric bone. This strengthens the rarity of CIB in our environment and in most populations.

Kumar et al found CIB in asterion (40.7%), occipitomastoid suture (6.15%), pterion (5.30%) and parietotemporal suture (4.42%). In the same study, there was no CIB within the sagittal, coronal sutures and bregma (Kumar et al., 2016). In our study, there was no CIB in the pterion, bregma and coronal suture but we found 4 sagittal CIB in 3 patients (Table 2).

Intrasutural bone may occur as a normal variant, but when the number is 10 or more it is said to be a marker of an underlying bony disease such as osteogenesis imperfecta, cleidocranial dysostosis, pycnodysostosis, congenital hypothyroidism and rickets (Marti et al., 2013). We found two autopsy subjects with at least CIB. These patients did not have any of the listed bony dysplasia or diseases.

In conclusion, CIB are common in adult Nigerian skulls. This should be taken into consideration by comparative and developmental anatomist, anthropologist, forensic physicians, neuroradiologists and neurological surgeons in population or patient evaluation and surgical planning.

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REFERENCES

1. Adejuwon SA, Olopade FE, Bolaji M (2013) Study of the location and morphology of the pterion in adult nigerian skulls. ISRN Anat, https://doi.org/10.5402/2013/403937
2. Bellary SS, Steinberg A, Mizayan N, Shirak M, Tubbs RS, Cohen-Gadol AA et al (2013) Wormian Bones- A Review. Clin Anat, 26(8): 922-927.
3. Bergman RA, Afifi AK, Miyauchi R (1988) Skeletal systems: Cranium. In: Compendium of human anatomical variations. Baltimore, Urban and Schwarzenberg, 197–205.

4. Cirpan S, Aksu F, Mas N (2015) The Incidence and Topographic Distribution of Sutures including Wormian in Human Skulls J. Craniofacial Surgery, 26(5): 1687-1690.
5. Cremin B, Goodman H, Spranger J, Beighton P (1982) Wormian Bones in Osteogenesis Imperfecta and other disorders. Skeletal Radiol, 8: 35-38
6. Das S, Suri R, Kapur V (2005) Anatomical observations on os inca and associated cranial deformities. Folia Morphol, 64:118-121.
7. Ersoy M, Evliyaoglu C, Bozkurt MC, Konuskan B, Tekdemir I, Keskil IS (2003) Epipteric bones in the pterion may be a surgical pitfall. Minim. Invasive Neurosurg, 46: 363–365.
8. Jeanty P, Silva SR, Turner C (2000) Prenatal diagnosis of wormian bones. J. Ultrasound Med, 19: 863–869.
9. Keskil S (2003) Common Surgical Pitfalls in the Skull. World Neurosurgery, 59(3): 229-231
10. Khan AA, Asari MA, Hassan A (2011) Unusual Presence of Wormian Sutural Bones in Human Skull. Folia Morphology, 70: 4
11. Kumar U, Prabha R (2016) Wormian Bones: Study on Dry Human Skulls in North Karnastaka Region. Int J Anat Res, 4(1): 1854-58
12. Malhotra VK, Tewari PS, Pandey SN, Tewari SP (1978) Interparietal bone. Acta Anat. (Basel), 101: 94–96.8.
13. Marti B, Sirinelli D, Maurin L, Carpentier E (2013) Wormian Bones in the General Paediatric Population. Diagnostic and Interventional Imaging, 94(4): 444-448
14. Matsumura G, Kida K, Ichikawa R, Kodama G (1991) Pterion and epipteric bones in Japanese adults and fetuses, with special reference to their formation and variations. Kaibogaku Zasshi, 66(5): 462-71.
15. Murphy T (1956) The pterion in Australian aborigine. American Journal of Physical Anthropology, 14(2): 225-244.
16. Natsis K, Piagkou M, Lazaridis N, Anastasopoulos N, Nousios G, Piagkos G, Loukas M (2019) Incidence, number and topography of Wormian bones in Greek adult dry skulls. Folia Morphol (Warsz), 78(2): 359-370.
17. Nayak SB (2008) Multiple Wormian Bones at the Lambdoid Suture in an Indian Skull. Neuroanatomy, 7: 52-53
18. Oguz O, Sanli SG, Bozkir MG, Soames RW (2004) The pterion in Turkish male skulls. Surg Radiol Anat, 26(3): 220-224.
19. Pal GP (1987) Variations of the interparietal bone in man. J. Anat, 152: 205-208.
20. Pedro A. Sanchez-Lara, John M. Graham Jr, Anne V. Hing, John Lee, Michael Cunningham (2007) The Morphogenesis of Wormian Bones: A Study of Craniosynostosis and Purposeful Cranial Deformation. AJMG, 143A(24): 3243-3251
21. Sanchez-Lara PA, Graham JM, Hing AV, Lee J, Cunningham M (2007) The Morphogenesis of Wormian Bones: A Study of Craniosynostosis and Purposeful Cranial Deformation. AJMG, 143A(24): 3243-3251
22. Saxena SK, Chowdhary DS, Jain SP (1986) Interparietal bones in Nigerian skulls. J. Anat, 144: 235–237.
23. Saxena SK, Jain SP, Chowdary DS (1988) A comparative Study of Pterion Formation and its Variations in the Skulls of Nigerians and Indians. Anthropol Anz, 46: 75-82
24. Seema PV, Mahajan A (2014) Human skull with complete metopic suture and multiple sutural bones at lambdoid suture – a case report. International Journal of Anatomical Variations, 7: 7–9.
25. Siddiqui AU, Gandhi KR, Daimi SRH (2013) A unique presentation of parietal notch bone with coexisting multiple wormian bones and complete metopism. International Journal of Biomedical Research, 04(10): 571-573.
26. Ukoha U, Oranusi CK, Okafor JI, Udemezue OO, Anyabolu AE, Nwamarachi TC (2013) Anatomic study of the pterion in Nigerian dry human skulls. Niger J Clin Pract, 16: 325-328