Original article

Morphological and topographical study of Wormian bones in cadaver dry skulls

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Abstract

Introduction: The Wormian bones are formations associated with insufficient rate of suture closure and regarded as epigenetic and hypostotic traits. It was reported that there exists racial variability among the incidence of these bones. In the present study, the aims were to find the incidence of Wormian bones in Indian skulls and to analyze them topographically. Material and methods: The study included 78 human adult dry skulls of Indian population which were obtained from the neuroanatomy laboratory of our institution. They were macroscopically observed for the incidence and topographical distribution of the Wormian bones. Results: The Wormian bones were observed in 57 skulls (73.1%) of our series. Remaining 21 skulls (26.9%) didn't show these variant bones. They were observed at the lambdoid suture in 56.4% cases (44 skulls; 14-bilateral; 18-right side; 12-left side), at the asterion in 17.9% (14 skulls; 3-bilateral; 2-right side; 9-left side), at the pterion in 11.5% (9 skulls; 4-right side; 5-left side), at the coronal suture in 1.3% (only one skull) and at the sagittal suture in 1.3% cases (only one skull). Conclusion: The current study observed Wormian bones in 73.1% of the cases from Indian population. This incidence rate is slightly higher compared to other reports and may be due to racial variations. These variant bones were more frequently seen at the lambdoid suture and were rare at the coronal and sagittal sutures. We believe that the knowledge of Wormian bones is of importance to the neuroanatomists, neurosurgeons, radiologists, anthropologists and morphologists.

Keywords: coronal suture, lambdoid suture, morphology, sagittal suture, Wormian bones.

1 Introduction

Wormian bones (WBs) are accessory bones which occur within the cranial suture and fontanelles, most commonly in the posterior sutures (SANCHEZ-LARA, GRAHAM JUNIOR, HING et al., 2007). They are named after the Danish anatomist, Olaus Wormius, who described them in a letter to Thomas Bartholin in 1643 (PENDERGRASS and SCHAEFFER, 1956; PRYLES and KHAN, 1979). However, WBs had been described in the past, with the first description attributed to Paracelsus (1460 to 1541 CE) (PRYLES and KHAN, 1979; PARKER, 1905). The alternative names of WBs include Schaltknochen (LEICHNER-WEIL, 1964), intercalary, sutural and intrasutural bones. These bones are detached portions of the primary ossification centres of the adjacent membrane bones (CREMIN, GOODMAN, SPRANGER et al., 1982). It was opined that they can be found in healthy individuals (BURGENER and KORMANO, 1997), however a higher incidence of multiple WBs have been found in a variety of congenital disorders like osteogenesis imperfecta, cretinism (hypothyroidism), cleidocranial dysostosis, progeria, hypophosphatasia, rickets etc. (BURGENER and KORMANO, 1997). The morphological knowledge of WBs is important in the diagnosis of these disorders (CREMIN, GOODMAN, SPRANGER et al., 1982). It was reported that their incidence is well suited for comparative studies as an anthropological marker or an indicator of population distance (GUMUSBURUN, SEVIM, KATKICI et al., 1997). Their knowledge is of interest to the human anatomy, physical anthropology and forensic medicine imaging (HERNANDEZ and ECHEVERRIA, 2009). Regarding their incidence, differences exist among various ethnic groups, with the highest incidence in Chinese individuals, as high as 80% (JEANTY, SILVA and TURNER, 2000). Ethnic variation in WBs may suggest possible genetic influences, but environmental influences could also play a role. Though the occurrence of WBs is quiet common, the observational data on them are poorly reported. At an orthopaedic meeting a question was asked concerning the nature and significance of WBs (CREMIN, GOODMAN, SPRANGER et al., 1982). But there was difficulty of giving an exact answer and it was debated that the literature about these bones is very scarce (CREMIN, GOODMAN, SPRANGER et al., 1982). The current study aimed to report the incidence of WBs in Indian skulls along with their topographical distribution. The morphological and clinical importances were emphasized with relevant review of literature.

2 Materials and methods

The study included a total number of 78 human adult dry skulls of Indian population. The specimens were obtained from the neuroanatomy laboratory of Manipal University. The exact ages and sex of the skulls were not determined. The skulls with vault removed and disarticulated were not included in the current study. The skulls were studied for the incidence and topographical distribution of the WBs. The

interparietal and preinterparietal bones which were seen at the lambda region were not considered as Wormian bones.

3 Results

The WBs were observed in 57 of the examined skulls (73.1%) and in the remaining 21 skulls (26.9%) they were not found. Most of the skulls showed WBs at multiple sites. They were observed at the lambdoid suture (Figure 1a) in 56.4% cases (44 skulls; 14-bilateral; 18-right side; 12-left side), at the asterion (Figure 1b) in 17.9% (14 skulls; 3-bilateral; 2-right side; 9-left side), at the pterion (Figure 1c) in 11.5% (9 skulls; 4-right side; 5-left side), at the coronal suture (Figure 1d) in 1.3% (only one skull) and at the sagittal suture (Figure 1e) in 1.3% (only one skull) of the cases. The WBs at the bregma was not observed in any of the skulls. The topographical distribution and frequencies of the WBs are summarized in Table 1.

4 Discussion

Wormian bones are formations associated with insufficient rate of suture closure and regarded as "epigenetic" and "hypostotic" traits (BARBERINI, BRUNER, CARTOLARI et al., 2008). They articulate with the surrounding bones by sutures, the dentations of which are more complex on the external side than on the internal side of the skull. The shape of these bones may be round, oval, oblong, triangular, quadrilateral or polygonal and vary in size from less than a millimeter in diameter to the one measuring 5 by 9 cm (PARKER, 1905). Parker (1905) mentioned several synonyms that were used, as follows, according to the discoverer: ossicula Andernaci, ossa Goethiano, according to shape: ossa triquetra, ossa triangularis, ossa quadratum, according to localization: suturaux, fontanellaires, insules, intercalaria, raphogeminantia, apicis, according to function: complementaria, ossa accessorii. It was suggested that WBs are not under the direct genetic control, but instead represent secondary sutural characteristics which are brought about by stress (BENNETT, 2005). These bones occur most commonly at the lambdoid sutures and within fontanelles. The etiology of WBs appears to relate to variations in the dural growth stretch along open sutures and within fontanelles causing ossification defects. Such sutural bones persist and are not incorporated into the adjacent bone during mineralization and maturation. With reference to their function Cruveilhier (1851) says:

> [...] They are looked upon as supplementary points of ossification and not as playing an important role in the mechanism of the solidity of the cranium, as would be supposed from the name, 'Keys of the vault,' or 'Clefts de route,' given them by some anatomists.

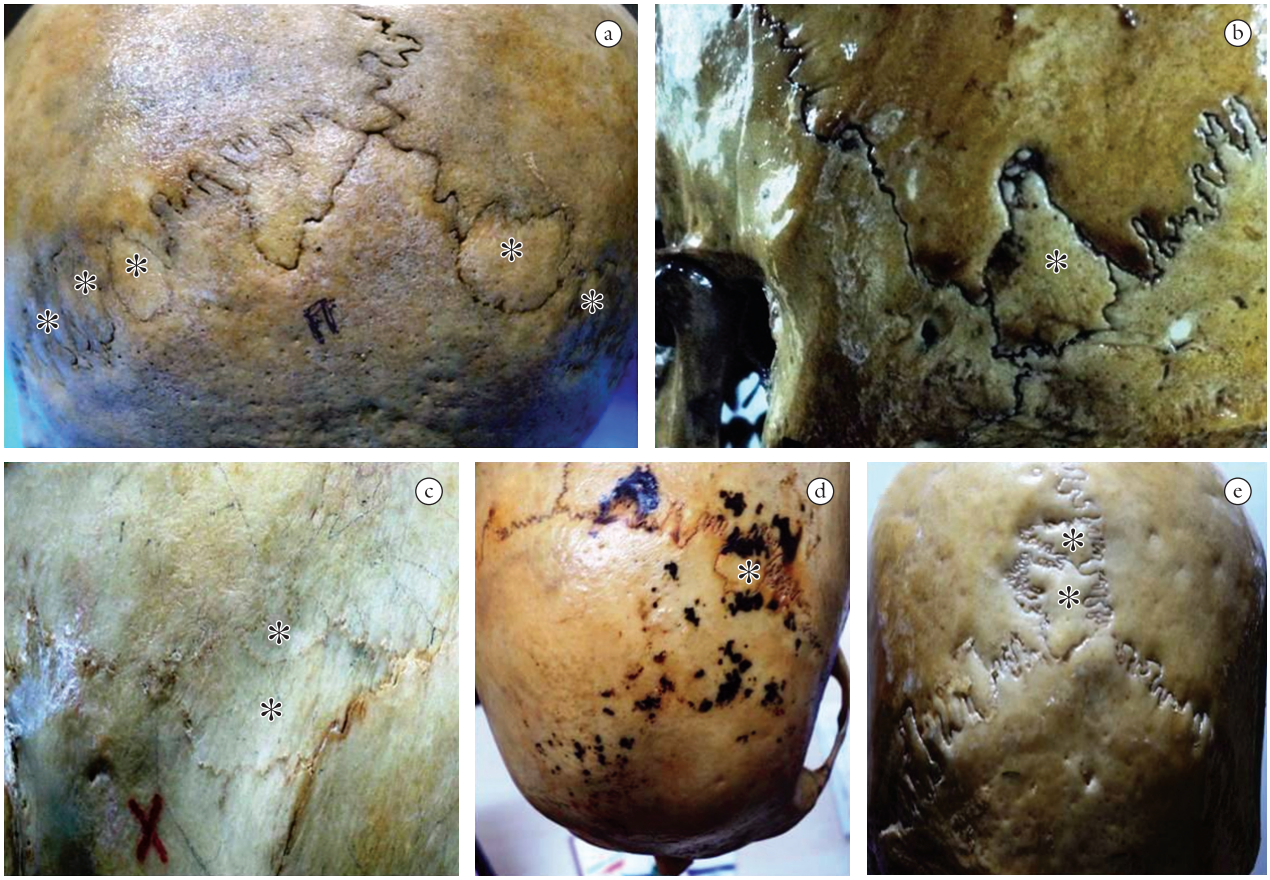

Figure 1. a) The skull showing multiple WBs at the lambdoid suture (*); b) The skull showing a single large Wormian bone at the asterion (*); c) The skull showing WBs at the pterion (*); d) The skull showing a single Wormian bone at the coronal suture (*); e) The skull showing WBs at the sagittal suture (*).

Table 1. Topographical distribution and frequency of the WBs.

Wormian bone present	Number and frequency
At lambdoid suture	44 (56.4%)
At asterion	14 (17.9%)
At pterion	9 (11.5%)
At coronal suture	1 (1.3%)
At sagittal suture	1(1.3%)
At bregma	nil (0%)

Table 2. Prevalence of WBs in different populations*.

Population	Prevalence (%)
Chinese	80.32
German	75
Australian	72.58
Iron Age/Romano-British	71.03
Melanesian	64.15
Lachish	63.41
Anglo-Saxon	55.56
Indian (present study)	73.1

*According to Brothwell (1963).

Skull growth is affected by dural attachments and is related to brain development. The developments of WBs occur at an early membranous stage of skull growth and it is difficult to correlate this with brain disorders or relate it specifically to any one type. It was reported that the WBs and skull growth are not affected by the mechanical distortion of the skull, which is a traditional practice in certain tribal communities (EL-NAJJAR and DAWSON, 1977).

Parker (1905) suggested that, the number of WBs increases with the capacity of the skull and a similar relationship exists with the total length of sutures, greater the sutural length, greater the number of WBs. Some authors (SANCHEZ-LARA, GRAHAM JUNIOR, HING et al., 2007) suggested that WBs may arise as a consequence of mechanical factors that spread sutures apart and affect dural strain within sutures and fontanelles. Since WBs belong to the neurocranium, they share its embryology. They appear as isolated ectopic islands of intramembranous ossifications. In the fetus, the diploe is not formed yet and thus WBs are composed of a single layer of compact bone on the dural side (KAPLAN and KEMP, 1991). Although the mechanisms of formation of WBs are unknown, some studies have shown that their presence may serve as a marker for the identification of anomalies of the central nervous system (JEANTY, SILVA and TURNER, 2000). When WBs occur as a normal variant, they tend to be smaller and less numerous than when they are associated with skeletal dysplasias (KAPLAN and KEMP, 1991). But most authors opine that they are not pathognomonic, as they occur in normal individuals, but the presence of more than ten sutural bones is unusual. It may warrant further investigations to identify an underlying pathology or hereditary disorder that has affected the skull growth at an early stage of development (CREMIN, GOODMAN, SPRANGER et al., 1982).

El-Najjar and Dawson (1977) suggested that the incidence is lower in fetuses (11.3%) than in adults (62.1% to 76.2%). Brothwell (1963) reported the prevalence of WBs among different populations and the data is represented in Table 2, which also includes the incidence rate of the current study. According to Bergman, Afifi and Miyauchi et al. (1988), about 40% of skulls have sutural bones at the vicinity of lambdoid suture. In the present study, the lambdoid suture showed the WBs in 56.4% of the cases. This is slightly more than the descriptions of Bergman, Afifi and Miyauchi et al. (1988). It was reported that, the second most common site of occurrence (about 25%) is in the coronal suture and the rest occur in any remaining sutures and fontanelles (CHAMBELLAN, 1883). Some authors reported that the second most common is the epipteric bone (pterion ossicle) found near the former anterolateral fontanelle (NAYAK, 2006). It was also reported that the incidence of epipteric bone is high in Indians. In the present study, the different topography was observed as the second common location was the asterion (17.9%) and third common location was the pterion (11.5%). A study by Saxena, Jain and Chowdhary (1988), also observed the similar incidence of epipteric bone (11.79%). The coronal and sagittal sutures showed very less prevalence (1.3%). The WBs at bregma were not observed in the current study.

It was reported that the presence of WBs at the pterion may lead to complications in making burr holes at the pterion (ERSOY, EVLIYAOGLU, BOZKURT et al., 2003). By simulating normal sutures or fractures, the occurrence of WBs may occasionally cause confusion during the radiological examination of the skull (PARKER, 1905). Sometimes, the WBs look like fractures and may confuse the radiologist or surgeon. It is more problematic if the fracture of skull is misinterpreted as a Wormian bone and the patient may lose the appropriate treatment at a right time. Hence the basic knowledge about these accessory bones is important for the doctors in day to day clinical practice.

In conclusion, the current study reports, 73.1% as the incidence rate of WBs in Indian skulls. This is slightly higher compared to other reports and may be because racial variations seem to exist. The WBs were more frequent at the lambdoid suture and less common at the coronal and sagittal sutures. We believe that the present study has provided additional information on the morphology and topography of the WBs. The clinical importances of these variant bones were emphasized with relevant review of literature. The knowledge of WBs is enlightening for the neuroanatomists, neurosurgeons, orthopedicians, radiologists, anthropologists and morphologists.

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