

## A LARGE SUPERNUMERARY BONE AT THE BREGMA AND METOPIISM CO-OCCURRING IN THE SKULL OF AN ANCIENT ROMAN IN SERBIA

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*Abstract* - An unusual anatomical variation was observed on a skull excavated at Lanište (Raška district, Serbia), a Roman necropolis dating back to the second half of the 4<sup>th</sup> century AD. The skull of an adult male showed a remarkably large supernumerary bone at the bregma co-occurring with a continuous persistent metopic suture. Few similar cases have been reported in scientific literature. We describe the case and discuss possible mechanisms and underlying causes, including pathological conditions.

*Key words:* bregmatic bone; metopism; Roman necropolis; 4<sup>th</sup> century AD.

### INTRODUCTION

Wormian bones (ossicles, supernumerary bones) represent one of the most frequently reported epigenetic variants in human skulls (Bergman et al., 1988). Located within the cranial sutures and fontanelles, these ossicles may be formed from a detached portion of the primary ossification centers of neurocranial bones, or they may rise from a new, abnormal ossification center. Their incidence can be related to a variety of pathological conditions (osteogenesis imperfecta, hypothyroidism, cleidocranial dysostosis, rickets etc.), but they are also commonly found in healthy individuals (Murlimanju et al., 2011). Some, such as wormian bones in the vicinity of the lambdoid suture, are frequent in human populations (40-50%), but others are very rare (Bergman et al., 1988; Brasili et al., 1999). One of these rare variations is a supernumerary bone placed at the bregma, the meeting point of the sagittal and coronal sutures; it forms within a large anterior fontanelle, situated be-

tween the anterosuperior angles of parietals and the superior angles of the separated halves of the frontal bone. The anterior fontanelle is the largest, kite-shaped, and it closes during the middle of the second year of postnatal life; if there are ossicles (one or more) at the bregma, they often reflect the shape of this fontanelle. The fusion of the metopic suture usually takes place during the first year, but completion of this process can last until an individual is 8 years old (Scheuer and Black, 2004). However, in some individuals, this suture remains unfused, even in adult life, and in such cases the term used is the persistent metopic suture (metopism).

The anterior fontanelle bone can relatively rarely be found in infant clinical cases (Agrawal et al., 2006; Carter and Anslow, 2009; Woods and Johnson, 2010). In adults, a number of bregmatic bone case studies from the forensic context have been reported in recent years (Nayak, 2006; Barberini et al., 2008; Hussain et al., 2010). Metopism is not as rare a vari-

ant, but its co-occurrence with a bone at the bregma is very rare. Here, we report a case of a large bregmatic bone co-occurring with metopism in an individual from a site in southwestern Serbia. We explore different causative mechanisms for this rare variant, including mechanical stress, genetic mutations and pathological conditions.

## MATERIALS AND METHODS

Lanište is located on the fluvial terrace of the river Ibar, near Baljevac (Raška district, Republic of Serbia; Fig. 1.). The archaeological team from the National Museum Kraljevo discovered 13 ancient Roman skeletal burials during 2001 and 2002. Coins and other grave goods date the burials to 4<sup>th</sup> century AD, most of them to the second half of the century (Bogosavljević-Petrović, 2003; Spasić, 2005). Apart from the cranium presented in this report (abbreviated L12 and showed in Fig. 2), the burial No. 12 contained only a damaged right radius, which could possibly belong to the same individual. The incompleteness of the skeletal material may be due to taphonomic factors, ancient, as well as the recent devastations, since many illegal excavation pits (made by unprofessional individuals searching for archaeological items made of metal) were recorded on the site. Considering the stratigraphic position of the remains and the fact that all inhumated burials on the site date roughly to 4<sup>th</sup> century AD, it is highly likely that the L12 skull also dates from this period. The skull was in a relatively good state of preservation, with only minor damage to its base and postmortem loss of teeth (dM<sup>1</sup>, dM<sup>2</sup>, sP<sup>4</sup>, sM<sup>1</sup> and sM<sup>2</sup> were preserved). We examined morphological characteristics of the skull in order to determine the sex (following Ferembach et al., 1980), and dental attrition (following Brothwell, 1981 and Lovejoy, 1985) in order to determine the age of the individual. The skull was examined by naked eye, and measured using sliding/spreading calipers.

## RESULTS

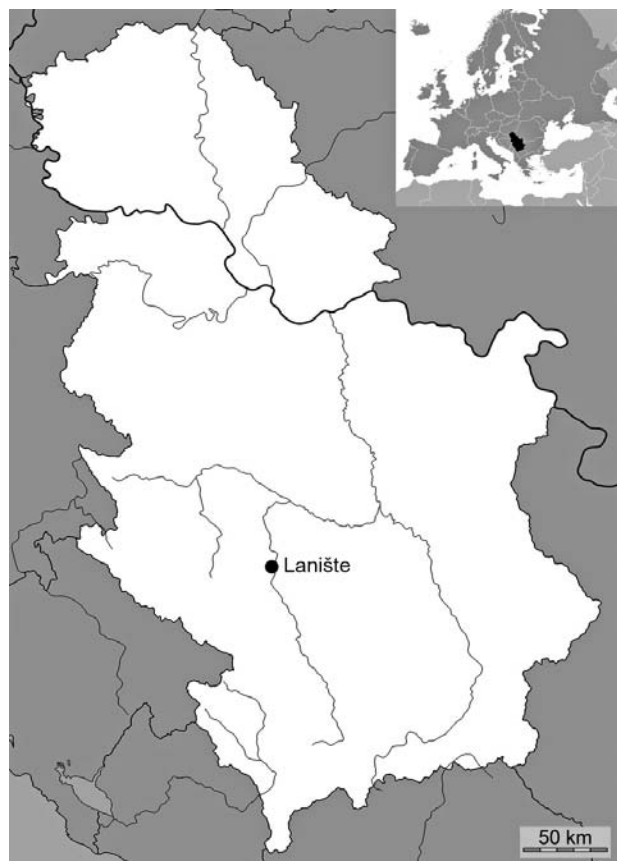
Our analysis showed that the skull belonged to an adult male individual. Since we had only a small

**Table 1.** Measurements of L12 skull according to Bass (1987) and Martin and Saller (1957) (marked with star).

Measurement/Index	L12
Max. cranial length (gl-op)	180
Max. cranial breadth (eu-eu)	157
Horizontal circumference (gl-op)*	554
Min. frontal breadth (ft-ft)	105
Interorbital breadth (d-d)*	23
Upper facial breadth (fmt-fmt)*	108
Upper facial height (n-pr)	68
Bizygomatic breadth (zy-zy)	142
Cranial index	87.22
Fronto-parietal index	66.87
Orbital index (right/left)	89.61
Nasal index	49.52
Maxilloalveolar index	121.65

number of skeletal individuals from the site for seriation procedures, we were not able to determine the specific dental attrition rate for the Lanište population. Considering this, the age of 35 to 45 years, based on the methods of Brothwell (1981) and Lovejoy (1985), do not reflect the exact age of the individual, but rather indicate only a broad adult age category. Further examination showed *rotatio dentis* (sP<sup>4</sup>), M<sup>3</sup> hypodontia and marked deviation of the nasal septum (Fig. 2). The neurocranium exhibited marked frontal and parietal curvatures. A cranial index over 85 (see Table 1) indicated that this is a hyperbrachycranial skull (Bass 1987: 69); some relevant measurements and indices are shown in Table 1 (following Bass, 1987 and Martin und Saller, 1957)

Wormian bones were noted on the left (20 x 20 mm) and right (25 x 17 mm) halves of the lambdoid suture and at the right asterion (15 x 13 mm) (Fig. 2); there were also few smaller lambdoid ossicles (now missing), but postdepositional distortion and damage impeded us from taking measurements. The large bregmatic bone was nearly pentagonal in form, reflecting the shape of the anterior fontanelle; the anterior margins of the bone were regular, straight, compared to the posterior, which showed a less regular outline (Figs. 2, 3). Measurements of the bone



**Fig. 1.** Site location map for the Ancient Roman cemetery at Lanište, with the position of Serbia in Europe.

were taken on the external table. The sagittal length of the bone was 69 mm; transverse breadth was 56 mm. Right anterolateral margin was 39 mm long, left was 42 mm; right posterolateral was 31 mm, left was 33 mm long, and the length of the posterior margin was 41 mm. At the endocranial aspect and in the vicinity of the bone, we observed small arachnoid calvarial defects (Kaufman et al., 1997). A continuous metopic suture was present, and the linear chord length from nasion to the most anterior point of the bregmatic bone was 96 mm.

#### DISCUSSION

According to Hauser and DeStefano (1989), the presence of a supernumerary bone at the bregma (*ossiculum fonticuli anterioris* or the bregmatic bone) is a

very rare occurrence in modern humans. In their survey, most of the examined skeletal populations (10 of 24) did not show any presence of this trait. For the populations where its presence had been recorded, frequencies were very low (0.2–2.5%), and there was no particular sex prevalence. O’Loughlin’s (2004) survey (which included 167 deformed and normal skulls) also detected no supernumerary bones at the bregma. This phenomenon had also been noted in other mammalian species, most notably in North American lynx (*Lynx rufus*), with high frequencies (37.5–44%). Bregmatic bones were also found in smaller mammal species (*Erinaceus europaeus*, *Rattus rattus*, *Sciurus vulgaris*, *Oryctolagus cuniculus*, and different species of *Microtus*), at very low frequencies, comparable to those found in humans. The shapes and dimensions of the bregmatic bones of these animals showed considerable intra- and interspecific variation (Manville, 1959; Pucek, 1962). Persistent metopic suture is not as rare as the occurrence of a bregmatic bone; according to Bergman et al. (1988), metopism occurs at frequencies ranging from 8.7% to 1% in recent populations. To our knowledge, only one other published case (Nayak, 2006) shows co-occurrence of the bregmatic bone and persistent metopic suture; compared to L12, Nayak’s case showed a relatively small and less regularly shaped bregmatic bone. The case presented by Barberini et al. (2008) was very similar to L12, both in terms of shape (nearly pentagonal) and size (relatively large) of the bregmatic ossicle; however, it showed no metopism, which we observed in the Lanište case. Another published case (Hussain et al., 2010) showed a rather differently shaped bregmatic bone (tetragonal), with an asymmetrical relative position (left lateral margin represented a continuation of the sagittal suture); this was clearly unlike the pattern we observed in L12.

A bregmatic bone probably arises from an abnormal ossification center in the fibrous membrane at the anterior median fontanelle (Hussain et al., 2010). Direct observations of skull development in transgenic mouse embryos have shown a dual embryonic origin of mammalian skull bones (Morriss-Kay, 2001). The parietals have a mesodermal origin



**Fig. 2.** Cranial projections of Lanište 12 skull.

and the frontal bone originates from the neural crest (Merrill et al., 2006); either of these two embryonic tissues could be responsible for the formation of a bregmatic bone, since it is positioned in the area of their direct interaction.

Sanchez-Lara et al. (2009) showed that “mechanical factors that spread sutures apart and affect dural

strain within sutures and fontanelles” play an important role in the formation of wormians within sutures or fontanelles. Their formation acts as a kind of adaptive response of the developing skull to mechanical stress (Oostra et al., 2005). These mechanical forces could be the result of different pathologies, cultural practice (purposeful cranial deformation) or of premature ossification of the cranial sutures (cranio-



**Fig. 3.** Detail view of the bregmatic bone, also illustrating the persistent metopic suture (arrow).

synostosis). The association of the bone formation in the anterior fontanelle with craniosynostosis was confirmed in clinical studies of children (Woods and Johnson, 2010; Agrawal et al., 2006). Since we see no evidence of craniosynostosis or artificial deformation in the L12 individual, other sources of mechanical stress could be involved. It is known that craniosynostosis causes skull shape deformations due to the limited growth of the skull bones in the direction perpendicular to the obliterated suture, which is compensated by growth in other directions (Aufderheide et al., 1998). Similarly, the peculiar shape of the L12 skull, exhibiting strong frontal/parietal curvatures (see Fig. 2) and hyperbrachycrany, could be related to the formation of the bregmatic bone. The growth of a large bone within the anterior fontanelle could act as a factor that modifies the development of the

surrounding neurocranial bones, frontals and parietals become “pushed” outwards, which is reflected in their bulging and brachycrany. Recent studies have found a positive correlation between brachycrany and both the formation of wormian bones (Sanchez-Lara et al., 2009) and metopism (Castilho et al., 2006).

In the context of pathological syndromes, the anterior fontanelle is a rare site for supernumerary bone formation. Woods and Johnson (2010) report association with acrocallosal syndrome (ACLS) in one patient; Elson et al. (1991) also report this association in one patient who, interestingly, also had a large, unfused anterior fontanelle at 3 years of age. Could this possibly account for the features we observed in L12? ACLS is a rare autosomal recessive disease, which is characterized by the absence of corpus callosum and severe mental retardation. An expected life span for individuals with ACLS has not been suggested yet, but Hodgson et al. (2009) believe that, with proper medical support, these individuals could survive well into adulthood. Skeletal anomalies are present in about 50% of cases, and the most frequent findings in this regard are pre- and post-axial polydactyly; among craniofacial anomalies, most frequent are a prominent forehead, macrocephaly (horizontal circumference  $>2$  standard deviations) and hypertelorism (Turolla et al., 1990). The forehead of L12 does not seem to be abnormally prominent; we detected no macrocephaly (see Table 1 for horizontal circumference), nor hypertelorism, since the interorbital distance measurement (Table 1) is normal for an adult male (Currarino and Silverman, 1960). Since the postcranium was not recovered, we cannot test the eventual presence of axial anomalies and therefore, we cannot confirm the (unlikely) hypothesis of ACLS to account for the features we observed in the L12 case.

Regarding metopism, some authors proposed that this trait could be an atavism, a reversion to an evolutionary primitive state (del Sol et al., 1989). However, although the fusion of a metopic suture has been widely accepted as a derived trait for anthropoids, Rosenberg and Pagano (2008) found that

many non-anthropoid fossil and living primates also showed frontal fusion. This could mean that this trait is not synapomorphic for anthropoids, and that, consequently, metopism could not be an atavism in humans. Other potential causes for metopism were also suggested (mechanical causes, hormonal dysfunction, etc.), but it seems clear that some genetic influence is probably involved (del Sol et al., 1989). This is in accordance with the studies of Torgersen (1951), which showed that there could be a hereditary basis for the metopism. There are no relevant hereditary studies concerning bregmatic bone, but this is mainly because the trait is so rare in humans. As Barberini et al. (2008) noted, it seems probable that some non-adaptive, direct genetic influence could be responsible for the development of a large bregmatic bone. If we accept the genetic basis for metopism and bregmatic bone formation, the rarity of their co-occurrence could imply that they involve different sets of genes.

### CONCLUSION

An adult male skull from Lanište, showing the co-occurrence of a large bregmatic bone and metopism, represents an important comparative specimen, which adds to the small number of published cases of this rare neurocranial variant. Different causes have been suggested for these phenomena, but their true nature remains poorly understood. Further research in different scientific fields is needed to decipher the underlying causes. A detailed genetic and long-term developmental study of patients diagnosed with bregmatic bone and/or metopism could prove to be particularly useful in this regard. Although a large bone at the bregma is usually considered a normal anatomical variant, the fact that it also occurs in some rare pathological conditions indicates the need to explore further the relationship of this phenomena with disease.

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### REFERENCES

- Agrawal, D., Steinbok, P. and Cochrane, D.D. (2006). Pseudoclosure of anterior fontanelle by wormian bone in isolated sagittal craniosynostosis. *Pediatr. Neurosurg.* **42**, 135-127.
- Aufderheide, A.C., Rodrigues-Martin, C. and Langsjoen, O. (1998). *The Cambridge Encyclopaedia of Human Paleopathology*. University Press, Cambridge.
- Barberini, F., Bruner, E., Cartolari, R., Franchitto, G., Heyn, R., Ricci, F. and Manzi, G. (2008). An unusually wide human bregmatic Wormian bone: anatomy, tomographic description and possible significance. *Surg. Radiol. Anat.* **30**, 683-687.
- Bass, W. M. (1987). *Human Osteology, A Laboratory and Field Manual*, Columbia: Missouri Archaeological Society.
- Bergman, R.A., Afifi, A.K. and Miyauchi, R. (1988). *Compendium of Human Anatomic Variation: Catalog, Atlas and World Literature*. Urban & Schwarzenberg, Baltimore and Munich.
- Bogoslavjević-Petrović, V. (2003). Struktura antičkog stanovništva u dolini Ibra. In *Rad Dragoslava Srejovića na istraživanju antičke arheologije* (Ed. Tasić, N.), 191-209. Memorijal Dragoslava Srejovića, Beograd.
- Brasili, P., Zaccagni, L. and Gualdi-Russo, E. (1999). Scoring of nonmetric cranial traits: A population study. *J. Anat.* **195**, 551-562.
- Brothwell, D. R. (1981). *Digging up bones* (3rd Ed.). Cornell University Press, Ithaca, NY.
- Carter, R. and Anslow, P. (2009). Imaging of the calvarium. *Semin. Ultrasound CT.* **30** (6), 465-91.
- Castilho, S.M.A., Oda, Y.J. and Santana, G.D.M. (2006). Metopism in Adult Skulls from Southern Brazil. *Int. J. Morphol.* **24** (1), 61-66.
- Currarino, G. and Silverman, F.N. (1960). Orbital hypotelorism, arhinencephaly and trigonocephaly. *Radiology* **74**, 206.
- Del Sol, M., Binignat, O., Bolini, P.D.A. and Prates, J.C. (1989). Metopismo no individuo Brasileiro. *Rev. Paul. Med.* **107** (2), 105-107.
- Elson, E., Perveen, R., Donnai, D., Wall, S. and Black, G.C. (2002). De novo GLI3 mutation in acrocallosal syndrome: broad-

- ening the phenotypic spectrum of GLI3 defects and overlap with murine models. *J. Med. Genet.* **39**, 804-806.
- Ferembach, D., Schwidetzky, I. and Stloukal, M. (1980). Recommendations for age and sex diagnosis of skeletons. *J. Hum. Evol.* **7**, 517-549.
- Hauser, G. and DeStefano, G.F. (1989). *Epigenetic variants of the human skull*. Schweizerbart, Stuttgart.
- Hodgson, B.D., Davies, L. and Gonsales, C.D. (2009). Acrocallosal Syndrome: A Case Report and Literature Survey. *J. Dent. Child.* **76** (2), 170-177.
- Hussain, S.S., Mavishetter, G.F., Thomas, S.T. and Muralidhar, P. (2010). Unusual sutural bone at Bregma – A case report. *Biomed. Res.* **21** (4), 369-370.
- Kaufman, M. H., Whitaker, D. and McTavish, J. (1997). Differential diagnosis of holes in the calvarium: Application of modern clinical data to palaeopathology. *J. Archaeol. Sci.* Vol **24** (3), 193-218.
- Lovejoy, C.O. (1985). Dental Wear in the Libben Population: Its functional pattern and role in the determination of adult skeletal age at death. *Am. J. Phys. Anthropol.* **68** (1), 47-56.
- Manville, R.H. (1959). Pragmatic bones in North American lynx. *Science* **130** (3384), 1254.
- Martin, R., und Saller, K. (1957). *Lehrbuch der Artropologie in systematischer Darsetzung*. G. Fischer Verlag, Stuttgart.
- Merrill, A.E., Bochukova, E.G., Brugger, S.M., Ishii, M., Pilz, D.T., Wall, S.A., Lyons, K.M., Wilkie, A.O.M. and Maxson, R.E. Jr. (2006). Cell mixing at a neural crest-mesoderm boundary and deficient ephrin-Eph signalling in the pathogenesis of craniosynostosis. *Hum. Mol. Genet.* **15**, 1319-1328.
- Morriss-Kay, G.M. (2001). Derivation of the mammalian skull vault. *J. Anat.* **199**, 143-151.
- Murlimanju, B.V., Prabhu, L.V., Ashraf, C.M., Kumar, C.G., Rai, R. and Maheshwari, C. (2011). Morphological and topographical study of Wormian bones in cadaver dry skulls. *J. Morph. Sci.* **28** (3), 176-179.
- Nayak, S.B. (2006). Presence of Wormian bones at bregma and paired frontal bone in an Indian skull. *Neuroanatomy* **5**, 42-43.
- O'Loughlin, V.D. (2004). Effects of different kinds of cranial deformation on the incidence of Wormian bones. *Am. J. Phys. Anthropol.* **123**, 146-155.
- Oostra, R.-J., van der Wolk, S., Maas, M. and Hennekam, R.C.M. (2005). Malformations of the axial skeleton in the museum Vrolik: II: craniosynostoses and suture-related conditions. *Am. J. Med. Genet.* **136A**, 327-342.
- Pucek, Z. (1962). The occurrence of wormian bones (*ossicula vormiana*) in some mammals. *Acta Theriol.* **6** (3), 33-51.
- Rosenberg, A.L. and Pagano, A.S. (2008). Frontal Fusion: Collapse of another anthropoid synapomorphy. *Anat. Rec.* **291**, 308-317.
- Sanchez-Lara, P.A., Graham Jr.JM., Hing, A.V., Lee, J. and Cunningham, M. (2009). The morphogenesis of wormian bones: A study of craniosynostosis and purposeful cranial deformation. *Am. J. Med. Genet.* **143A**, 3243-3251.
- Scheuer, L., and Black, S. (2004). *The Juvenile Skeleton*. Elsevier Academic Press, San Diego.
- Spasić, S. (2005). Nalazi novca na antičkoj nekropoli Lanište kod Baljevca. *Naša Prošlost* **6**, 117-138.
- Torgersen, J. (1951). The developmental genetics and evolutionary meaning of the metopic suture. *Am. J. Phys. Anthropol.* **9**, 98-102; 193-210.
- Turolla, L., Clementi, M. and Tenconi, R. (1990). How wide is the clinical spectrum of the acrocallosal syndrome? Report of a mild case. *J. Med. Genet.* **27**, 516-518.
- Woods, R.H. and Johnson, D. (2010). Absence of the anterior fontanelle due to a fontanellar bone. *J. Craniofac. Surg.* **21**(2), 448-449.

