JOURNAL OF FORENSIC SCIENCES

J Forensic Sci, May 2011, Vol. 56, No. 3
doi: 10.1111/j.1556-4029.2011.01718.x
Available online at: onlinelibrary.wiley.com

CASE REPORT

ANTHROPOLOGY

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Positive Identification by a Skull with Multiple Epigenetic Traits and Abnormal Structure of the Neurocranium, Viscerocranium, and the Skeleton*

ABSTRACT: Wormian bones are small ossicles appearing within the cranial sutures in more than 40% of skulls, most commonly at the lambdoid suture and pterion. During the skeletal analysis of an unidentified male war victim, we observed multiple wormian bones and a patent metopic suture. Additionally, the right elbow was deformed, probably as a consequence of an old trauma. The skull was analyzed by cranial measurements and computerized tomography, revealing the presence of cranial deformities including hyperbrachicrania, localized reduction in hemispheral widths, increased cranial capacity, and sclerosis of the viscerocranium. Besides unique anatomical features and their anthropological value, such skeletal abnormalities also have a forensic value as the evidence to support the final identification of the victim.

KEYWORDS: forensic science, skull, cranial sutures, identification, wormian bones, metopism

Morphological alterations in the skeleton have been described as a useful tool in forensic identification (1,2). Wormian bones (sutural bones and sutural ossicles) are small, irregular ossicles, varying in size and number, which appear within the cranial sutures. Wormian bones and patent sutures are referred to as "epigenetic traits," or intrinsically innocuous minor skeletal variations of the human skull (3). Most commonly they are located at the lambdoid suture (4) and the pterion (5). The appearance of wormian bones is related to various genetic factors (6,7) including metabolic disorders, inheritable syndromes (4,8), and environmental factors such as mechanical stress (7,9–11), or a combination of genetic and environmental factors as in craniosynostosis (10). Mechanical stress may induce the appearance of wormian bones and other epigenetic traits, in particular when applied in the early years of development, either in the form of external pressure, as seen in artificially deformed crania from New World archaeological sites (10) or in the form of internal pressure of the growing brain (12). Mechanical stress seems to be related to the appearance of wormian bones specifically at the lambdoid area (10). On the other hand, there are suggestions that epigenetic traits are under greater genetic than environmental influence (11). A clear mechanism whereby genetic factors contribute to the formation of wormian bones is unknown. It is considered that they originate from small ossification centers that appear within the sutures and fontanels (13,14). Some metabolic and inheritable disorders have also been connected to the appearance of wormian bones (4,8), but in more than 40% of the skulls, they appear as an isolated finding (15).

Here, we report a case of a unique skull discovered during the exhumation of human skeletal war remains characterized by the appearance of multiple epigenetic traits such as wormian bones in the lambdoid area, unilateral pterionic wormian bone, and a patent metopic suture, accompanied by deformities in the facial skeleton, the occipital bone, and the right elbow. We also address on the application of such specific findings in forensic identification.

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*Supported by the grants from the Croatian Ministry of Science, Education and Sports (108-0000000-0030 and 108-1080229-0140).

Received 5 Nov. 2009; and in revised form 6 April 2010; accepted 25 April 2010.

Case Report

In the autumn of 2006, from the exhumations of the 1991–1995 Croatian Homeland war (16) in the village Smoljanac, in Licko-Senjska County, a solitary burial place located near an abandoned cottage was disinterred. The location of the burial place was reported by the residents of the village, the deceased's neighbors. Inside the burial place, a single skeleton of a middle-aged man was found. In addition to well-preserved skeletal remains, clothing and boots were found, wrapped in a blanket along with the remains.

Visual Examination

At general inspection, the skull appeared deformed (Fig. 1*A*). The rest of the skeleton was normal in macroscopical appearance and size. Upon thorough visual examination of the skull, multiple

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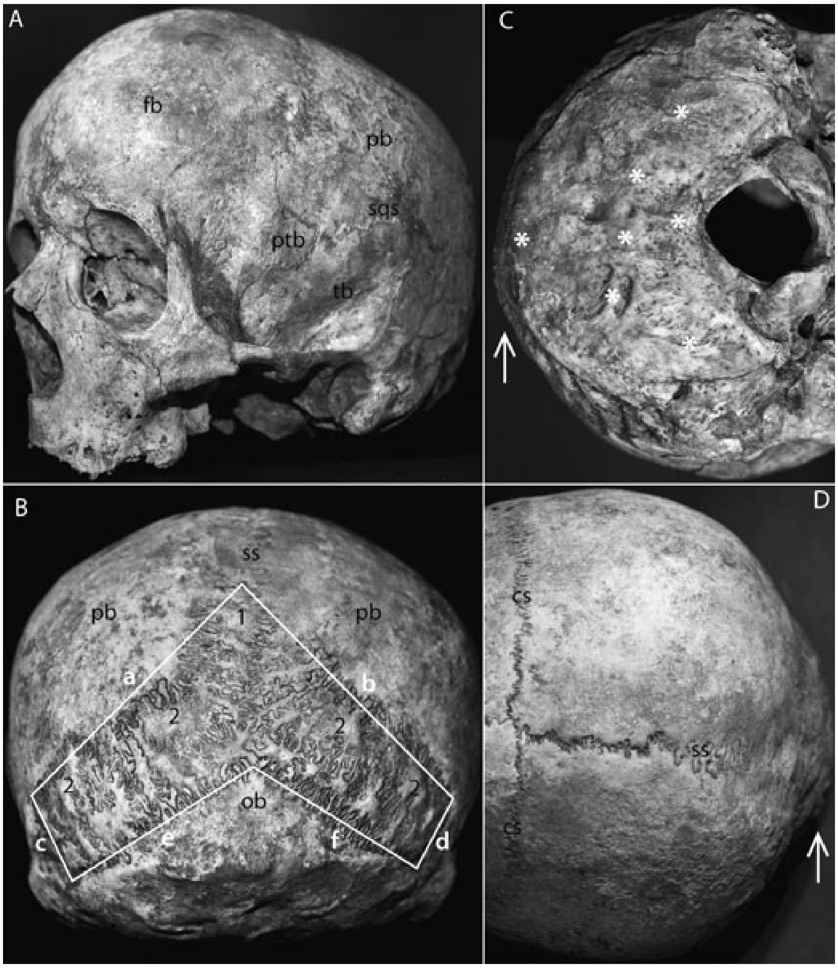

FIG. 1—*Deformed appearance of the skull, multiple wormian bones in the region of lambda, deformations of the occipital squama, and the position of muscle insertions on the occipital bone. (A) Left anterolateral view of the skull; (B) posterior view; (C) inferior view; (D) superior view. a, Left anterolateral margin, 99 mm; b, the right anterolateral margin, 96 mm; c, left lateral margin, 34 mm; d, right lateral margin, 28 mm; e, left posterolateral margin, 77 mm; f, right posterolateral margin 74 mm; cs, coronal suture; fb, frontal bone; ob, occipital bone; pb, parietal bone; ptb, pterionic bone; sqs, squamous suture; ss, sagittal suture; tb, temporal bone; 1, apical wormian bone; 2, lamboid wormian bones; arrow, conical bulging of the occipital squama; asterisks, insertion points of the neck and back muscles to the occipital bone.*

wormian bones were noted, most appearing in the area around the lambdoid suture, and, additionally, left pterionic wormian bone. The bones varied in shape and size and were partially fused at isolated sites. There were a total of 31 clearly visible wormian bones. According to their location (10), there was one pterionic, one parietomastoid, one parietal notch, one apical ossicle, and 27 lambdoid ossicles. The ossicles were situated in the area between the parietomastoid suture and the lambda, separating the parietal bones from the squama of the occipital bone. They formed an irregular lambdoid-shaped hexagonal area with two anterolateral, two lateral, and two posterolateral margins (Fig. 1*B*). The occipital bone was asymmetric, with a prominent conical bulging on the left portion of the bone, closely above the inferior nuchal line (Fig. 1*C*,*D*). The squamous portion of the occipital bone appeared relatively small in comparison with the basilar part of the bone.

In the area of the left pterion, we observed a rectangular pterionic wormian bone (Fig. 2*A*). Sutures surrounding this bone were visible on the external osseous table. The sutures along the frontal, the parietal, and the temporal bone were parts of sphenofrontal, coronal, and squamous sutures, respectively. The suture along the sphenoid bone was less patent and less visible than the other sutures (Fig. 2*A*).

The squama of the frontal bone was divided by a patent metopic suture spreading from bregma to nasion (Fig. 2*B*). The upper end of the metopic suture reached the coronal suture slightly to the right of the midline. Additionally, a bony mass, measuring 1.1 cm in diameter, corresponding to an osteoma, was noted on the left portion of the squama of the frontal bone (Fig. 2*B*).

Coronal, sagittal, squamous, and lambdoid sutures were patent, with increased digitations in the posterior part of the sagittal suture, from bregma to lambda (Fig. 1*B*,*D*), and at the lateral parts of the coronal sutures (Fig. 2*A*).

Cranial Measurements

Cranial measurements were performed to confirm the presence of cranial deformities (17), and the results are summarized in Table 1.

The comparison of these results with the values for craniofacial variations in the Croatian population (18) revealed decreased maximum head length (MHL) (186 mm in our case vs. 194 ± 6.69 mm mean), bizygomatic breadth (129 mm in our case vs. 141 ± 6.78 mm mean), and minimal frontal breadth (97 mm in our case vs. 110.05 ± 6.34). Head index (HI, calculated according to

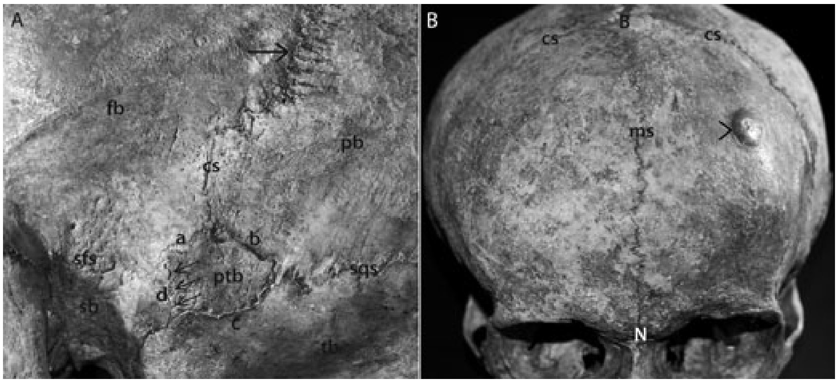

FIG. 2—*Pterionic wormian bone and the patent metopic suture. (A) Left lateral view of the skull showing the pterionic bone. (B) Anterosuperior view showing the patent metopic suture. Small arrows, newly formed suture between the sphenoid and the wormian bone; large arrow, increased digitations of the lateral part of the coronal suture; arrowhead, a bony mass corresponding to an osteoma; a, frontal margin, 16 mm; b, parietal margin, 16 mm; c, temporal margin 19 mm; d, sphenoid margin, 11 mm; B, bregma; N, nasion; cs, coronal suture; fb, frontal bone; ms, metopic suture; pb, parietal bone; ptb, pterionic bone; sb, sphenoid bone; sfs, sphenofrontal suture; sqs, squamous suture; tb, temporal bone.*

TABLE 1—*Cranial measurements of the skull.*

Cranial Measurement*	Description	Value (mm)
Maximum head length	Glabella–opisthocranion	186
Maximum head breadth (MHB)	Euryon–euryon	163
Bizygomatic breadth	Zygion–zygion	129
Auricular height (AH)	Meatus acusticus externus–vertex	129
Frontal breadth	Frontotemporale–frontotemporale	97

*The measurements were taken according to Buikstra and Ubelaker (17).

the formula HI = MHB × 100/MHL) was 87.63, which pointed to hyperbrachycrania (range 85.5–90.9; [18]). These results show that, when compared with normal skulls, this skull was shorter, but wider, and with a narrow forehead and face.

The overall cranial capacity (CC) was calculated by the Lee and Pearson's formula for men [CC = 0.000337 (MHL − 11 mm) (MHB − 11 mm) (AH − 11 mm) + 406.01] (19). Eleven millimeter subtractions, used to account for skin and fat tissue around the skull, were excluded from the calculation because they were not present in our case. The CC was 1724.02 cm^3, revealing an increase when compared with the results of Niyazi et al., where the mean CC for men was 1411.64 ± 118.9 cm^3 (19), and Rushton, where the mean CC for Caucasoid men was 1419–1445 cm^3 (20).

Computerized Tomography (CT)

The skull was scanned with a 16-slice MSCT scanner (Somatom Sensation 16; Siemens, Erlanghen, Germany) at 5-mm slices with a 1.0-mm reconstruction increment. The 4-mm multiplanar image reconstructions were performed at the axial, coronal, and sagittal plane at workstation Leonardo (Siemens). For the purpose of viewing, measuring, and plotting, we used Adobe Illustrator CS4 (Adobe Systems, Inc., San Jose, CA), Adobe Photoshop CS4, and medical imaging viewing station Siemens SIENET MagicView-300 VA40A software.

On the scans, the skull appeared asymmetric, with alterations in the bone structure dominating on the left side. The walls of the left maxillary sinus were thickened and sclerotic, particularly in the anterior and the lateral wall, reducing the size of the sinus. Maximum width of the sinus was 4.1 cm on the right side and 3.3 cm on the left side (Fig. 3). Furthermore, the sclerotic changes were

FIG. 3—*Reduction in the left maxillary sinus maximal width. Horizontal section of the skull at the level of the sella turcica. ms, maxillary sinus.*

also present in the left zygomatic bone, left half of the frontal bone, sphenoid bone, and posterior ethmoid sinus (Figs 3 and 4). To determine the effect of sclerosis and the deformities at the lambdoid suture on the hemispheral width, we used the method described by Barberini et al. (13). Briefly, a CT scan image of the lowest available horizontal section, which excluded the size of the left frontal sinus cavity, was analyzed. Fourteen equally distanced points were placed on the line connecting the frontal and occipital internal protuberance, and the distances from the marked points to the internal table were measured on both the right and the left side. These values were then used to calculate the right/left (R/L) hemispherical width ratio. The hemispherical width in the frontal portion of the cranium was reduced on the left side; in the temporal portion, the ratio was close to 1, whereas in the occipital portion, the reduction in the hemispherical width was noted on the right side (Fig. 4, Table 2).

As severe deformities of the occipital bone resulted in the flat appearance of the posterior part of the skull base, indicative of platybasia, we measured the sphenoid angle on the CT scan of the

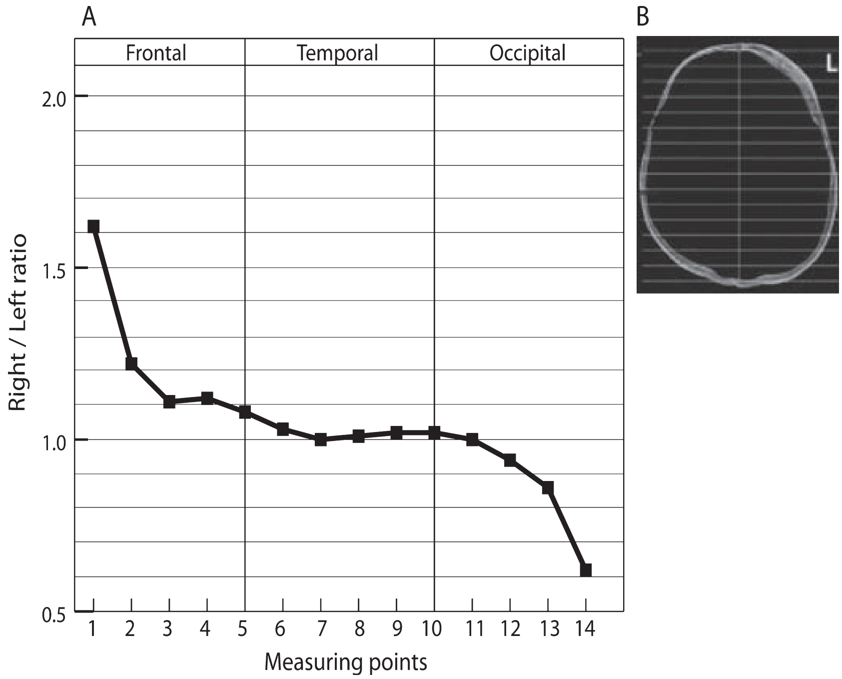

FIG. 4—*Reduction in the anterior left hemispherical width and the posterior right hemispheral width. (A) The ratio between the right and the left hemispherical width in the frontal, temporal, and occipital parts of the skull; (B) computerized tomography image of the lowest available horizontal section of the skull, which excluded the size of the left frontal sinus cavity, showing 14 points of measurement.*

TABLE 2—*Hemispherical widths through the cranium and the calculated right/left (R/L) ratio.*

		Hemispherical Width (cm)		
Portion	Measuring Points*	Right	Left	R/L Ratio
Frontal	1	4.52	2.79	1.62
	2	5.42	4.45	1.22
	3	5.90	5.31	1.11
	4	6.55	5.84	1.12
	5	7.13	6.60	1.08
Temporal	6	7.30	7.1	1.03
	7	7.44	7.44	1.00
	8	7.60	7.55	1.01
	9	7.67	7.50	1.02
	10	7.55	7.40	1.02
Occipital	11	7.23	7.23	1.00
	12	6.44	6.88	0.94
	13	5.41	6.28	0.86
	14	3.10	4.95	0.62

*The measurements were taken according to Barberini et al. (13).

sagittal plane at the level of sella turcica, according to the method described by Koenigsberg et al. (21). The sphenoid angle was 125°, falling in the normal range (119 ± 9°; [22]).

Right Elbow Deformity as a Result of an Old Trauma

The rest of the skeleton had no apparent morphological anomalies except at the right elbow, which had a severely altered morphology (Fig. 5), most likely because of untreated healing of earlier trauma. The radius was shifted distally and laterally, and a new pseudo-articulation was formed between the damaged humerus and the ulna. The ulna was rotated laterally, and its coronoid process and the olecranon formed the lateral articular surface, while the lateral portion of the damaged trochlea of the humerus formed the medial surface. The lateral distal part of the right humerus was avulsed, with consequent absence of the lateral epicondyle and the capitulum. The cubital parts of the radius and the ulna contained massive bony projections indicating increased deposition of new bone in the area of the trauma. As a result, the movements in the elbow joint were limited, occurring only in the new pseudo-articulation, and the forearm was nearly fixed in incomplete supination, combined with partial lateral rotation in the shoulder joint.

Discussion

The appearance of wormian bones is under genetic (6,7,14) and/or environmental (7,9,10,14) influence and, as an isolated finding occurs in 14% of population (14). Furthermore, appearance of wormian bones is associated with various congenital anomalies and disorders of the central nervous system (8).

The most likely explanation for the features of this skull is the influence of both genetic and environmental factors, with forces generated by the muscles of the neck and back, and the pressure from increased intracranial pressure (ICP) being the environmental factors. The influence of genetic factors is likely because of the appearance of multiple traits. Which specific genes were involved was not determined, but the nature of deformation of the occipital bone suggests developmental abnormalities. Abnormal genetic influence may have caused abnormal formation of the occipital bone, by which early ossification centers did not fuse at the proper time, resulting in the appearance of multiple wormian bones at the lambdoid area, an abnormal small size of the occipital squama, and thus, also abnormal positioning of the insertion points of the muscles of the neck and back, which insert at the area between the nuchal lines and at the nuchal ligament. This further resulted in altered distribution of mechanical forces of these muscles, which may have promoted the appearance of additional wormians. The increased CC suggests the existence of increased ICP, which may have also played a role in the appearance of additional wormians.

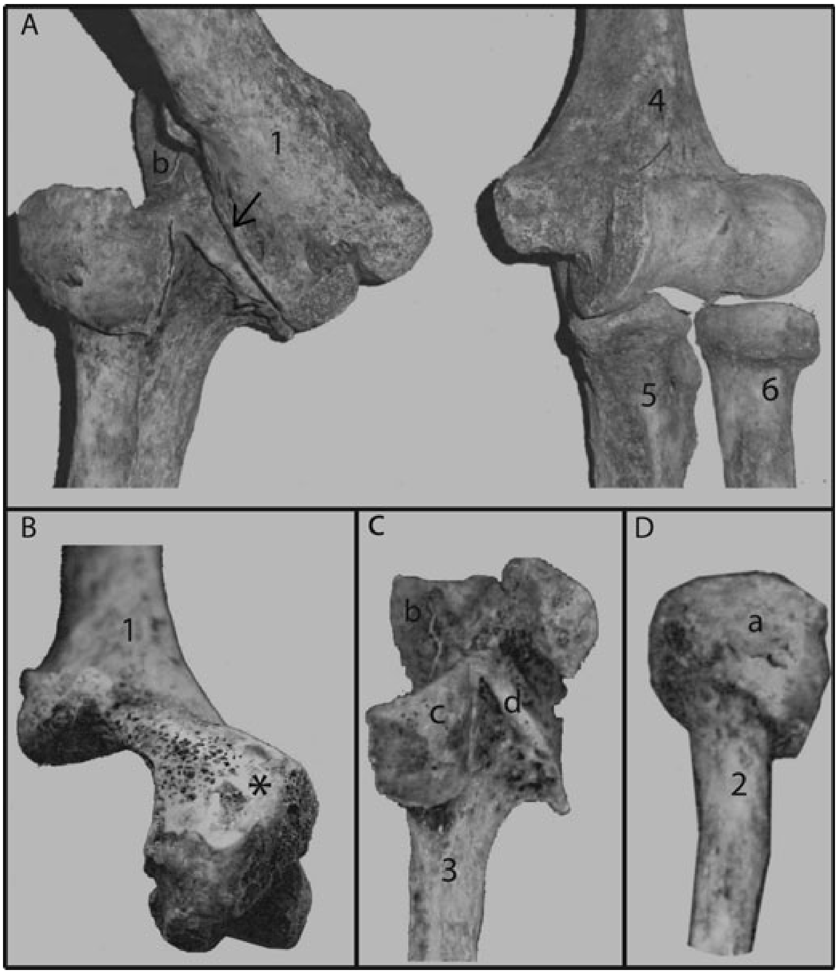

FIG. 5—*The right and the left elbow joint, and the articular surfaces of the right elbow. (A) Posterior view of the right elbow joint showing the new bone arrangement after the healing of the untreated trauma and the posterior view of the left elbow joint with normal anatomy; (B) damaged distal part of the right humerus, with avulsed lateral part of the articular surface; (C) the proximal part of the right ulna; (D) the proximal part of the right radius, with prominent bone depositions; 1, right humerus; 2, right radius; 3, right ulna; 4, left humerus; 5, left ulna; 6, left radius; asterisk, the avulsed part of the humerus, the medial face of the new articulation between the right humerus and the right ulna; arrow, the new articulation between the right humerus and ulna; a, head of the radius; b, new bone deposition at the proximal part of the ulna; c, bone deposition at the proximal part of the ulna where the head of the radius fused with the ulna; d, the lateral face of the new articulation between the right humerus and the ulna.*

Increased CC was shown to be associated with the presence of wormian bones (4,14) and their appearance may serve as an adaptation method of the cranium to larger cranial volume (12,14). Fragmentation of the lambdoid area with multiple wormian bones contributed to the flattened appearance of the occipital portion of the skull base and the prominent conical bulging of the occipital squama. Future research on the influence of increased CC and ICP, and the mechanical forces generated by the muscles of the neck and the back which insert at the occipital bone, on the appearance of wormian bones is needed to confirm or reject this hypothesis.

The complexity of the formation of the calvarial bones and of cranial sutures (23–27) plays a role in the formation of sutural bones. The occipital squama ossifies in two ways. The planum occipitale (interparietal part) is formed by membranous ossification, whereas the planum nuchale (supraoccipital part) by cartilaginous ossification (25). The membranous part of the occipital squama ossifies from two primary interparietal ossification centers (one at each side), two secondary interparietal centers, and in some cases, additional preinterparietal centers (23,27), which fuse with each other and unify with the cartilaginous part of the bone by the end of fetal development. The union of the squama with the lateral and the basilar part of the occipital bone occurs by 6 years of age (15). Failure of these centers to fuse may result in the appearance of small sutural bones or larger interparietal (inca) bones (28).

The frontal bone is formed by membranous ossification from two primary ossification centers above the supraorbital margins at the end of second gestational month (15). Failure of the fusion of two primary ossification centers of the frontal bones by the age of 8 years results in metopism (15), which occurs in 1–10% of skulls (29). Metopic suture present in the case study skull had no visible discontinuation from nasion to bregma, although the internal table appeared intact on the CT scans. Metopism is known to be related to the influence of genetic rather than the environmental factors (11). Metopism is commonly associated with brachycrania and increased CC (30), which were both present in the case study skull, and may further be associated with other anomalies of the cranium, or underdevelopment or absence of the clavicle (5). In the case study skull, the upper end of the metopic suture reached the coronal suture slightly to the right of the midline. Such asymmetry is often described in metopic skulls and is attributed to the greater development of the left hemisphere (30).

Besides neurocranial anomalies, the viscerocranium was also severely affected. In addition to the frontal bone, hyperosseous alterations were also noted in the left zygomatic and the maxillary bone. The anatomical structure of the affected bones was normal, pointing to the fact that these alterations have formed after the development of the viscerocranium has been completed and corresponded to a mild form of fibrous dysplasia, a genetic ossification disorder. The narrowing of the right frontal and the left occipital portion of the skull, present in our case, may cause various neurological symptoms, such as headache, facial pain, vertigo, neuralgias, vision and hearing loss, as well as personality and cognitive disorders, and as such have to be considered in differential diagnosis (31). The untreated left elbow trauma resulted in a unique clinical presentation, which could be very useful in forensic identification.

Many congenital disorders and anomalies may result in a phenotype described in our case, including ossification disorders and disorders of the mesoderm, such as osteogenesis imperfecta, Menkes syndrome, Prader–Willi syndrome, osteopetrosis, hydrocephalus, hypothyroidism, craniosynostosis, and others (4,8). Taking into account the multiple characters of the skull anomalies, an inheritable disorder is the probable cause of the phenotype described in our case, although the exact diagnosis could not be established. The bones of the left elbow had massive bony depositions, pointing to increased bone formation in response to trauma, which may also fit into the clinical presentation of a generalized ossification disorder. The trauma was estimated to have occurred after the growth of the bones had been completed. A more accurate estimation cannot be made based solely on the visual examination.

The disappearance of this person was reported by his neighbors, who filed a missing person report in 1991. No living relatives could be identified, which made DNA comparison impossible (16). The age indicated in the report was 52, and the missing person was described as having a large, disproportional head, odd appearance of his left hand, as well as some degree of intellectual disability. Findings on the case study skeleton's skull and left elbow agreed closely with the description in the missing person report, including the age at death estimated during forensic identification. The morphological abnormalities of the viscerocranium and the neurocranium may have been related to the changes in the brain development and subsequent mental disability. The anthropological and morphological findings described in the case study and the location of the grave near the cottage where the missing person used to live played a key role leading to a positive identification of the deceased.

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