

CASE REPORT

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A rare case of the scaphocephalic skull of an adult male

Shahriar Ahmadpour*  and Khadijeh Foghi

Abstract

Background: Scaphocephaly is a craniofacial anomaly where the skull is disproportionately long and narrow. Premature closure of the sagittal suture is thought to be at the core of the etiopathology for scaphocephaly.

Case presentation: The skull is a well-preserved skull found in an antiquated graveyard at Parkand village Daregaz, Iran, with no visible signs of surgical manipulation. The craniofacial indices were measured. The maximum cranial length was 200 mm, and the measured length of nasion-bregma was 140mm, whereas most of the measurements were less than the normal scaphocephalic indices. Another morphological finding was the flattening of the frontal bone.

Conclusions: The cranial vault morphometry and morphology of the presented case fit the scalp. In addition to the characteristics of scaphocephaly, the presented skull was of a rare trait, frontal flattening, which has not been reported before.

Keywords: Scaphocephaly, Frontal flattening, Skull, Daregaz

Background

The skull is the most complex bony structure in the human body. It is composed of the neurocranium and viscerocranium bones which ossify in quite different modes. The calvarial bones are formed through intramembranous ossification. At the sutural sites of cranial bones, osteoprogenitor cells differentiate into bone-forming osteoblasts in a synchronous time-dependent manner (Luo et al. 2017). During post-natal life, sutures remain open and allow the harmonious growth of the skull and the developing brain (Morris-Kay and Wilkie 2005). Premature fusion or closure of one or more sutures, craniosynostosis, interrupts the coordinated growth of the skull and leads to morphological craniofacial anomalies. Craniosynostosis might be classified depending on the underlying mechanism, presence of other disorders, or number of fused sutures (Kajdic et al. 2018). Studies

have shown that the cumulative prevalence of craniosynostoses has increased significantly with no obvious cause (Cornelissen et al. 2016). It is thought both environmental factors (e.g., intrauterine fetal head constraint, abnormal position, oligohydramnios, prenatal exposures to teratogens, maternal smoking, and anti-epileptic drugs) and genes (single-gene mutations, chromosome abnormalities, and polygenic background) may all be predisposing factors for the craniosynostosis (Kajdic et al. 2018). Based on the typical features, various types of craniosynostoses have been described.

One of the most frequent forms of craniosynostosis is scaphocephaly (Captier et al. 2005). The term scaphocephaly refers to the condition where the skull is disproportionately long and narrow (Skrzat et al. 2014). Premature closure of the sagittal suture is thought to be at the core of the etiopathology for scaphocephaly (Brah et al. 2020). It is the most common form of isolated craniosynostosis, accounting for 40–60% of single-suture synostosis (Jane Jr et al. 2000; Thwin et al. 2015). Studies have provided evidence that craniosynostosis is associated with a higher risk of impaired cognitive

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development, neurological disorders, and psychological disturbances (Aldridge et al. 2017; Bellew and Chumas 2015; Adamo and Pollack 2010). Although nowadays, adults with dolichocephalic skulls are rarely observed because of advances in prenatal diagnosis and surgical interventions, the study of cranial morphology remains an active field of inquiry in bioarchaeology. The present case is an adult scaphocephalic skull with unique characteristics, frontal flattening, which has not been reported before. We measured the cranial indices and compare them with the previous reports.

Case presentation

The skull (without mandible) was found in an antiquated graveyard at Parkand village, 10 km from Daregaz, North Khorasan Province. Dargaz County is located in the northeastern Iranian Plateau, near to borders of Turkmenistan. The skull was found accidentally among the roots of a dried tree. Due to climatic erosion and drought, the roots of the tree were exposed. The skull was buried deeply among roots. There was no sign of a burial ceremony including shroud and weapons. Soil and dried roots were gently removed. Further extraction revealed no remains of the bony skeleton. Unfortunately, the skull chronology is unknown. Sexing was determined by the evaluation of the morphology of the bones of the skull. Prominent superciliary arches, well-developed temporal lines, and a large mastoid process outweigh in favor of a male character of the investigated skull (Nagare et al. 2018). The skull was gently cleaned with a soft brush and examined with a magnifier. There were no signs of sutural fusion with the exception of the anterior part of the sagittal suture. In this study, direct anthropometric

measurement techniques were taken by the researcher. Basic anthropological measurements were taken directly on the skull using appropriate instruments including flexible tape and a sliding caliper (Weber et al. 2008). Cranial measurements performed on the dolichocephalic skull are shown in Table 1. All anthropometric measurements were performed on the examined skull by three experts and then compared to the previously reported adult male dolichocephalic skulls (Skrzat 1999).

Results

The most striking features of the examined dolichocephalic skull were the extremely narrow and long cranium. In norma verticalis, the investigated skull showed half-fused sagittal sutures and non-obiterated coronal and lambdoid sutures. In turn, the frontal norma of the examined skull presented a highly sloped frontal squama, a long and narrow face (Figs. 1, 2, 3, and 4). The maximum cranial length ($g-op=200mm$) was 11.1mm longer than the mean normative value (188.9mm). The maximum cranial breadth ($eu-eu=120mm$) was significantly decreased in comparison with the normal dolichocephalic skulls ($eu-eu = 136.9mm$). The minimum frontal breadth ($ft-ft =80mm$) was 17mm smaller than the mean of normal dolichocephalic skulls ($ft-ft = 17mm$). The parietal chord ($b-l =100 mm$) was 17.8 mm shorter than the normal dolichocephalic skulls ($b-l =117.8mm$). The occipital chord ($l-o =80 mm$) was 17.5 mm shorter than the reference skulls ($l-o =97.5 mm$). The biggest difference was related to the frontal chord ($n-b = 140mm$) in comparison to the mean of the reference skulls (115mm). Both the arch length of ($b-l =116mm$) and arch length ($l-o =105 mm$) were smaller than the reported mean

Table 1 Measurements performed on scaphocephalic skull

Mesurment	Description
1. Maximum cranial length ($g-op$)	Direct distance measured between glabella and opisthocranion
2. Maximum cranial breadth ($eu-eu$)(Euryon)	Maximum width of skull perpendicular to midsagittal plane
3. Bizygomatic diameter ($zy-zy$)	Maximum breadth across the zygomatic arches, perpendicular to the median plane
4. Midfacial width ($zm-zm$)	The shortest distance between right and left zygomaxillare
5. Basion-bregma height ($ba-b$)	Direct distance from the lowest point on the anterior margin of foramen magnum to bregma
6. Upper facial height ($n pr$)	Direct distance from nasion to prosthion
7. Minimum frontal breadth ($ft-ft$)	Direct distance between 2 frontotemporale
8. Frontal chord ($n-b$)	Direct distance from nasion to bregma taken in the midsagittal plane.
9.Parietal chord ($b-l$)	Direct distance from bregma to lambda taken in the midsagittal plane
10. Occipital chord ($l-o$)	Direct distance from lambda to opisthion
11. Biasterionic width ($ast-ast$)	The shortest distance between right and left asterion
12.Arc length ($n-b$)	Arch length measured between nasion and bregma
13. Arc length ($b-l$)	Arch length measured between bregma and lambda
14. Arc length ($l-o$)	Arch length measured between lambda and opisthion



Fig. 1 Lateral view (Norma lateralis)

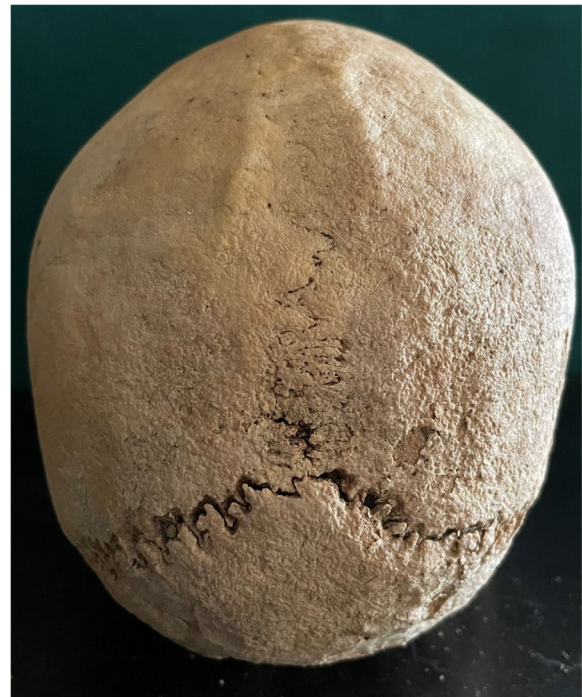


Fig. 3 Posterior view (Norma occipitalis)

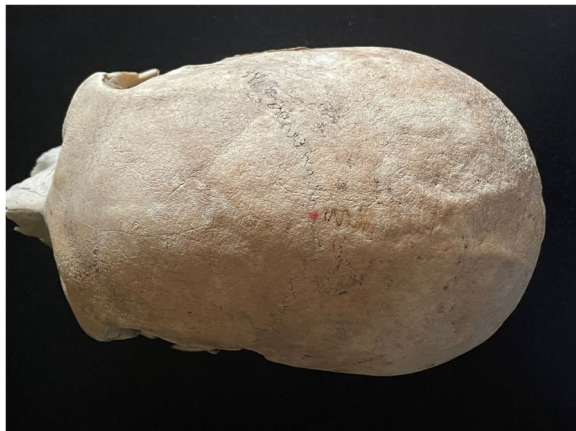


Fig. 2 Superior view (Norma verticalis)



Fig. 4 Anterior view (Norma frontalis)

values (117.8mm and 97.5mm, respectively). The arc length (n-b) was smaller than those of the mean normative value. The ratio between (ba-b) and maximum cranial breadth (eu-eu) was greater than the mean of the dolichocephalic skulls (125 vs 101, ratio 23.2). The bizygomatic diameter (zy-zy = 110mm) also revealed another significant contrast in comparison to the reference value (129.8mm) (Table 2).

Discussion

Craniosynostosis is a condition where premature closure of one or more cranial sutures results in abnormal craniofacial morphology as well as neurocognitive deficits (Heuzé et al. 2010). Craniosynostosis can occur primarily

and secondarily. It is believed that developmental error during embryogenesis leads to primary craniosynostosis whereas metabolic disorders such as hypothyroidism,

Table 2 Craniofacial measurements and indices of the scaphocephalic skull compared with reported dolichocephalic skulls of the male adults by Skrzat et al. (3)

Measurement and index	Case X	Mean Reference	X- ref. mean
g-op	200	188.9	+11.1
ba-b	135	139.2	-4.2
eu-eu	120	136.9	-16.9
ft-ft	80	97	-17
ast-ast	110	110.1	-0.1
n-b	140	115	+25
b-l	100	117.8	-17.8
l-o	80	97.5	-17.5
arc n-b	130	131.8	-1.8
arc b-l	116	131.4	-15.4
Arc l-o	105	120.2	-15.2
n-pr	70	69.3	+0.7
zy-zy	110	129.8	-19.8
zm-zm	90	96.3	-6.3
Ba-b/eu-eu x100	125	101.8	+23.2

The measurements and index of the case X. The difference between the case and reference(X-ref)

teratogens, and some genetic syndromes result in secondary craniosynostosis. More than 80% of primary craniosynostosis cases occur as an isolated condition (Kim et al. 2016). Scaphocephaly is the most common form of isolated craniosynostosis in which restriction of growth perpendicular to the sagittal suture results in an elongated and narrow cranium (Governale 2015). The examined scaphocephalic skull is of unknown historical and ethnical origin, and due to technical limitations, we were unable to take the advantage of radiometric dating technique. We believe the results of this study can be noticeable in archeological and biomedical aspects. First, from an archeological aspect, artificial modification of the human skull corresponds to the period between 45,000 B.C.E and 600 C.E (Khudaverdyan 2011).

Studies have shown that skull modification was a prevalent cultural practice in the prehistoric era. At least 27 skulls have been reliably defined as having been artificially modified in the West of Iran. It seems that the practice of artificial skull modification was most likely restricted to certain members of the community or even gender distinction as the skulls found in Ali Koš (Marcus 1993).

It is noteworthy to mention that Iranian prehistoric artificially modified skulls have been recovered from the West of Iran, while there are few documented studies about such practice in the northeast of Iran. Thus, any conclusion about the similarity between this case and previous reports should be taken cautiously. With respect

to the location of the buried skull, it may likely raise the sacrificial ritual issue or crime scene. According to the study, sacrificial ritual in ancient Iran was restricted to animals (Ghalekhani and Bushehri 2016), but at present, we could not eliminate the possibility of the criminal act. From a biomedical aspect, the craniofacial measurements and indices of the present skull provide an excellent example of scaphocephaly. The maximum cranial length of the present skull indicates the anteroposterior compensatory expansion that in turn may affect the length of the parietal and occipital bones. The parietal and occipital chords were smaller than those of reference scaphocephalic skulls, while the frontal chord was significantly larger than the reference value. Interestingly, the sagittal suture was semi-obiterated, while premature closure of the sagittal suture and coronal suture compensatory mechanism has been known as the main players of morphological changes in scaphocephaly (White et al. 2021). The presented scaphocephalic skull does not manifest frontal and occipital knobs which are the typical symptoms of scaphocephaly. Additionally, the maximum cranial breadth and viscerocranium breadth were smaller than the mean reference values (17mm and 16.9mm smaller than those of the reference, respectively). The results of this study are in line with the previous reports. For instance, Skrzat et al. (2014) reported a scaphocephalic skull with a maximum cranial length of 201 mm. In another study, Weber et al. (2008) measured 18 scaphocephalic skulls and the mean maximum cranial length was 203mm. The reported values are nearly close to our scaphocephalic skull. The development and growth of the skull is a complex process that is controlled and regulated by a wide variety of biological processes including ecology, locomotion, diet, behavior, climate, and genes (Chico 2011). For instance, studies have provided evidence that mutation in the Homeobox containing transcription factor *MSX2*, *FGF1,2,3*, and helix-loop-helix transcription factor *TWIST* leads to craniosynostosis (Rice et al. 2003). From an evolutionary perspective, it has been postulated that the number of skull bones follows a general trend toward the reduction in the number of individual parts, resulting from losses and fusion of bones (Williston's law) (Ascarrunz et al. 2019). Although the investigated skull presents a scaphocephalic skull, the most striking and unique feature is the frontal bone external shape that has not been reported in previous studies. Contrary to the normal scaphocephalic skulls, the present skull is of a flattened frontal bone (frontal flattening). This finding may be partially explained by artificial modification with hand-shaping (Bronfin 2001; Meikelejohn et al. 1992) or as a feature of a genetic disorder. Considering our technical limitations on the one hand and lack of any burial evidence on the

other hand, the distinction between artificial modification and other possible causes is infeasible. From an evolutionary perspective, frontal flattening is one of the most significant morphological features of mid-Pleistocene fossil hominids and Neanderthal when compared to the more vertical modern median-sagittal profile (Bookstein et al. 1999). Admittedly, we are unable to verify such an assumption because of the following reasons: first, the investigated skull is of unknown historical origin, and second, craniosynostosis is also caused by a metabolic disorder, hematologic disorder, and gene mutation (Lattanzi et al. 2017). Therefore, without radiometric dating and the medical history of the investigated skull, any conclusion should be taken cautiously. According to Virchow's law, malformation found in scaphocephaly includes elongation of anteroposterior (frontal-occipital) diameter and shortening of biparietal diameter (Chico 2011) as we reported here. Such dynamic changes in the skull would affect the external features of the skull bone particularly the frontal and occipital bones. Various forms of the frontal bone shape comprising bilateral and rectangular, normal, or semisphere have been reported in scaphocephaly (Chico 2011). Interestingly, the frontal bone of the examined skull presents another, flattened frontal bone, and variant. One of the possible explanations for the flattened frontal bone could be the semi-obliterated sagittal suture. In other words, premature obliteration of the anterior half of the sagittal suture interrupts the normal coordinated growth of the skull, leading to the shortened occipital chord (as measured in the present case) and finally compensatory anteroposterior elongation of the skull. This finding may accentuate the importance of the external feature of the frontal bone in neuroarcheologic studies as Bookstein's study showed. He compared the frontal bone profiles in Archaic and modern humans. The results of their study revealed substantial external differences between the Archaic and modern samples. These differences were mainly confined to the squamous part of the frontal bone (Bookstein et al. 1999). Accordingly, skull growth could affect the underlying developing brain and subsequently cognitive capabilities. It has been shown that craniosynostosis affects the cortical and subcortical structures (Shim et al. 2016). Additionally, premature closure of the cranial sutures may lead to abnormalities in the cerebral topography (Aldridge et al. 2017). For instance, the narrowing of the posterior area of the brain and constriction of the brain between the left Sylvian fissure and left Rolandic fissure have been reported by Aldridge et al. (2017). It has been also documented that sagittal synostosis is associated with an increased risk of Chiari malformation (Strahle et al. 2011). Furthermore, other studies have provided evidence that children with sagittal synostosis

demonstrate reading comprehension and language problems (Kapp-Simon et al. 2016; Shim et al. 2016). It should be noted that it would not be tenable to generalize such neurological disorders to all scaphocephalic cases. So we assume the investigated skull was the subject of our study belongs to a person with a healthy mental condition.

Conclusions

Here, we reported a scaphocephalic skull with frontal flattening which has been less noticed. The present case represents a relatively rare finding in anthropology and paleopathology, but these data must be interpreted with caution because of our technical and methodological limitations. With respect to the lack of skeleton remains as seen in this case, taking advantage of advanced technologies including genetic profile analysis, radiometric dating technique, imaging, and more archeologic excavation particularly in northeastern Iran is recommended.

Abbreviations

l-o: Arc length; b-l: Arc length; n-b: Arc length; ast-ast: Biasterionic width; ba-b: Basion-bregma height; zy-zy: Bizygomatic diameter; n-b: Frontal chord; g-op: Maximum cranial length; Euryon: Maximum cranial breadth (eu-eu); zm-zm: Midfacial width; ft-ft: Minimum frontal breadth; l-o: Occipital chord; b-l: Parietal chord; n-pr: Upper facial height.

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Authors' contributions

The co-author of this case presentation contributed to searching the materials and craniometry. The author(s) read and approved the final manuscript.

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Availability of data and materials

The authors confirm that the material of this study is available in the Anatomy Department.

Declarations

Ethics approval and consent to participate

NA

Consent for publication

We, the undersigned, give our consent for the publication of identifiable details, which can include photograph(s) and/or videos and/or case history and/or details within the text ("Material") to be published in EJFS. I confirm that I have seen and been given the opportunity to read both the material and the article (as attached) to be published by EJFS. We have discussed this consent form with Dr. Shahriar Ahmadpour who is the corresponding author of this paper. I understand that EJFS may be available in both print and on the internet and will be available to a broader audience through marketing channels and other third parties. Therefore, anyone can read material published in the Journal. I understand that readers may include not only medical professionals and scholarly researchers but also journalists and general members of the public.

Competing interests

The authors declare that they have no competing interests.

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