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Metopic skull with occipitalization of the atlas

S. Nikolova, D. Toneva, Metopism and atlas occipitalization

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ABSTRACT

In this study we report a combination of anatomic variations in the *neurocranium* of an adult male skull. The skull is scanned using an industrial µCT system Nikon XT H 225. The cranial vault shows a persistent metopic suture, a remnant from the mendosal suture and supernumerary bones. Cranial base inspection reveals atlas occipitalization (AO), basilar kyphosis, unusually shaped dorsum sellae and bilateral interclinoid bridging. AO is extensive without signs of atlantooccipital articulation. The anterior and posterior arches of the atlas and the right transverse process are fussed to the occipital bone above. The complete fusion of the posterior arch causes a formation of bone canals for the vertebral arteries passage into the cranium. AO in this case is not related to a reduction of the foramen magnum dimensions and *clivus* length.

Key words: persistent metopic suture, atlas assimilation, supernumerary calvarial bones, wormian bones, intraclinoid bridging, caroticoclinoid foramen, posterior interclinoid foramen

INTRODUCTION

Anatomic variations in the skull also called discrete, non-metric, discontinuous or epigenetic traits have been considered intrinsically innocuous minor skeletal variants [10, 14]. The boundaries between variations and anomalies, however, are not so clear. Fluctuation of size, form and position within a commonly experienced range has been considered as “normal variation”, whereas “anomaly” has been accepted as synonymous to structural abnormality caused by aberrant developmental processes. Unlike anomalies, variations have no impact on the function of a particular structure under normal circumstances, even though sometimes harmless variations could have a negative effect [33] and could indicate generalized disorders [6, 17]. Persistent sutures and supernumerary bones are variations in the cranial configuration causing an abnormal partition of the vault. *Metopism* arises when the metopic suture, which forms between the growing halves of the frontal bone after the 16th fetal week [8] and normally closes before the end of the first postnatal year [38], fails to fuse. Persistent metopic suture is found to occur with varying frequency in different population groups from being absent to exceeding 15% [9, 20, 39]. It has been observed that metopic crania differ from non-metopic ones in many aspects. Metopic crania have distinctive cranial morphology [26, 27, 29] with frequently manifested supernumerary calvarial bones, remnants from embryonic sutures [9, 21, 26, 35] and underdeveloped frontal sinus [23, 24]. They demonstrate a general delay in calvarial sutures closure [28] and a wide array of midline closure defects [37]. Moreover, persisting MS is a common finding in some congenital disorders related to impaired bone formation [6, 17, 19]. Supernumerary bones could arise due to non-fusion of normally present ossification centres or they may appear from additional ossification centres. The squamous part of the occipital bone is an intricate site where different variations arise due to faulty fusion between the ossification centres of the interparietal part of the occipital squama and a frequent occurrence of wormian bones [17, 18].

Atlas occipitalization (AO), also known as atlas assimilation, atlanto-occipital fusion, occipitocervical synostosis and atlantooccipital joint ankylosis is a spinal anomaly of cranial base characterized by a partial or complete fusion of the first cervical vertebra (C1, atlas) to the occipital bone. AO is a congenital osseous abnormality in the craniovertebral junction, which arises during the early embryonic period (4th fetal week) as a result of maldevelopment due to an incomplete segmentation of the first cervical sclerotome into cranial and caudal components. Then, the caudal portion of the fourth occipital somite fusses with the entire first cervical

sclerotome and the cranial portion of the second cervical sclerotome, resulting in an assimilation of the atlas into the occipital region [4]. Incomplete incorporation or failure of segmentation of the last occipital and C1 sclerotomes leads to a spectrum of fusion-related anomalies and accessory structures, many of which are asymptomatic. The occipitovertebral border is an embryologically unstable area and the phenomenon of cranial–caudal border shifting is still not entirely understood. A caudal shift in the position of the atlanto-occipital demarcation is more common than a cranial shift. A caudal shift causes an assimilation of the atlas into the occipital bone as well as a basilar impression/platybasia and presence of paracondylar and epitransverse processes. A cranial shift results in an occipital vertebra (proatlas) expressed by precondylar tubercle or process at the anterior border of foramen magnum, transverse basilar clefts, bipartite condylar facets and divided hypoglossal canals [7]. Paracondylar process has also been considered to be a manifestation of parts of the proatlas transverse process [12].

AO is normally congenital, but in rare cases it may be a result of diseases such as osteomyelitis, arthritis, syphilis, tuberculosis and other infections or traumatic injuries of the cervical vertebrae [31, 36]. Factors such as malnutrition or disease-related disturbances during development or genetic anomalies may also predispose a fetus toward developing AO [31]. It has been reported that AO could be associated with Pfeiffer, Crouzon and Apert syndromes involving craniosynostosis, as well as with Goldenhar, Klippel-Feil and Pierre-Robin syndromes [5, 16]. The fusion between the occipital bone and the atlas varies as it could be localized or extensive, uni- or bilateral. In majority of the cases, the fusion is localized to the region of the atlantooccipital articulations and the complete bony union is rare [4, 7]. Since AO and the related abnormalities could be asymptomatic they often goes undetected and AO is commonly diagnosed incidentally in radiographs, intraoperatively or during autopsy. The reported AO frequency ranges between 0.03 - 3.6 % in the contemporary populations [12, 16, 31].

In this study, we report a case of an adult male cranium showing a rare combination of different anatomical variations in the *neurocranium* including persistent metopic suture and AO. We also discuss the origin and possible clinical complications of these variations.

CASE REPORT

The investigated cranium (Fig. 1) is part of the osteological material stored in the Ossuary at the National Museum of Military History (Bulgaria). The cranium belonged to an

adult Bulgarian soldier who died in the wars at the beginning of the 20th century. The cranium was scanned using an industrial μCT system Nikon XT H 225 following the previously established optimal scanning protocol for dry skulls [25]. The sagittal suture closure was assessed on cross-sectional tomograms and regression models for age-at-death prediction, elaborated on the same population [25], were applied. Based on the sagittal suture closure degree, the age-at-death of the individual was assessed to be 34 years.

Variations in the calvarial morphology. The crania had a persistent metopic suture lying between *nasion* and *bregma* (Fig. 2a). Supernumerary calvarial bones and remnants from embryological sutures were observed. A rounded preinterparietal bone [18] along with a few separate wormian bones of different size occupied the upper triangular portion of the occipital squama (Fig. 2b). A remnant from the mendosal suture was visible on the left side of the occipital squama (Fig. 2b). An epipteric ossicle was found at the right pterion (Fig. 1c), which could be classified as “false” or “incomplete” epipteric bone (*os epiptericum spurium totum peritemporale*) according to the classification of Kadanoff and Mutafov [12].

Variations in the cranial base. The atlas was fused to the occipital bone (Fig. 2d). The posterior vertebral arch was complete and entirely fused to the occipital squama, whereas a small aperture (width – 9.2 mm; height 4.1 mm) between the anterior arch and the anterior margin of foramen magnum was observed (Fig. 3a). Both lateral masses were ossified to the occipital condyles without any sighs of atlantooccipital articulation between them. The inferior articular facets were asymmetrical. The transverse processes were normally developed, enclosing the transverse foramina. The left transverse process was free, while the right one was fused directly to the inferior surface of the jugular process of the occipital bone (Fig. 3b). This way, an additional transversal foramen was formed, enclosed by the fussed occipital bone and the transverse process of the atlas (Fig. 3b). At the level of the fused margins of the posterior arch and the occipital bone, right behind the fused lateral masses, bilaterally were formed bony canals for the vertebral arteries passage (Fig. 3b). Behind the right canal, there was a small vertical bony bridge between the occipital bone and the fused posterior arch forming a foramen (Fig. 3b). The hypoglossal canals on both sides were of normal appearance and position. The atlas midsagittal axis was slightly rotated to the left side. The atlas was also inclined from the transversal plane as its right side (fused C1 transverse process) was in an upper position compared to the free left side (Figs. 2d, 3a).

The intracranial inspection revealed an unusually shaped dorsum sellae along with erosion of its base and top (Fig. 4a). There was also a bilateral interclinoid bridging. According to the classification of Archana et al. [3], there was an interclinoid bridging Type II, mixed type on the right side (Fig. 4b). It was formed between the anterior, middle and posterior clinoid processes. The ossified ligaments enclosed an anterior interclinoid foramen (caroticoclinoid foramen) between anterior and middle clinoid processes, and a posterior interclinoid foramen between the middle and posterior clinoid processes (Fig. 4b). On the left side, there was an interclinoid bridging Type I, contact type [3] representing a bone bridge between the anterior and middle clinoid processes, enclosing an anterior interclinoid foramen (Fig. 4c). The cranial base angle was constructed and measured in the midsagittal plane between the landmarks *nasion*, *sellae* and *basion* (Fig. 5). The borderline values of cranial base angle delimiting a normal from flexed and extended base angle were used after Koenigsberg et al. [13]: basilar kyphosis, an extensive flexion of the skull base, CBA < 125°; normal angulation, CBA between 125°-143°; platybasia, an abnormal flattening of the skull base, CBA > 143°. The measured cranial base angle in this case indicated basilar kyphosis (119.52°).

Linear measurements: Cranial length (g-op) – 174 mm; Cranial width (eu-eu) – 135 mm; Cranial height (ba-b) – 130 mm; Maximum foramen magnum anteroposterior diameter (H) – 39.5 mm; Maximum foramen magnum width (W) – 35.6 mm; *clivus* length – 47.7 mm

Calculated indices: Cranial index (eu-eu/g-op) – 77.6, *mesocran*; Height-length index; (ba-b/g-op) – 74.7, *orthocran*; Height-breadth index (ba-b/eu-eu) – 96.3, *metriocran*.

The actual area of the foramen magnum was calculated according to the formula of Radinsky [32]:

Area = π × 1/4 × W × H, where W is the maximum foramen magnum width and H is its maximum anteroposterior diameter in the midsagittal plane. Foramen magnum area – 1103.9 mm^2.

DISCUSSION

The causes for non-fusion of the frontal bone halves are widely discussed but still not fully understood. *Metopism* has been attributed to various conditions and causes such as increased intracranial pressure, mechanical stress, endocrine dysfunction, growth retardation, mental defects, heredity and heredo-specific factors as well as to specific cranial deformations

such as plagiocephaly, stenocrotaphy, brachycephaly, scapho-cephaly and hydrocephaly [1, 35]. In this case, the cranium is of medium size without any obvious deformations in its shape and size. *Metopism* is frequently associated with underdevelopment of the frontal sinus [17, 19, 20, 23, 24]. However, in the reported case all paranasal sinuses are well-developed.

AO and the supernumerary bones in the occipital squama could be explained by developmental anomalies, which occur during the occipital bone formation and seem to be unrelated due to the different timetables and ossification patterns of the occipital bone partition in which they evolve. AO arises around the 4th week of gestation as a result of maldevelopment of the intrasegmental fissure of the C1 sclerotome segment, with a consequent lack of recombination of the C1 somite [4]. The interparietal part of the occipital squama is a triangular space situated between the highest nuchal line and both parietal bones, which undergoes an intramembranous ossification (after the 8th fetal week) from a variable number of ossification centres giving rise to numerous variations in cases of non-fusion. The presence of a true preinterparietal bone is still questionable, but we use this term to designate a single bone or a group of bones (bipartite, tripartite or multipartite preinterparietal bone) of different size forming a triangular territory in the central lambda region, and separated by a transverse suture located higher than the midline between the lambda and the highest nuchal line [18]. In the reported case, it is observed a relatively large rounded bone occupying the upper portion of the occipital squama. This bone together with a few smaller irregular wormian bones delineates a triangular space below *lambda*. Wormian bones could arise from a fragmentation in the primary ossification centres or they may develop from additional centres [17]. Besides the supernumerary bones in the upper portion of the occipital squama, there is a remnant from the mendosal suture on the left side, which lies at the level of the highest nuchal line i.e. on the demarcation line between the primary and secondary ossification centres. Thus, considering the occipital bone development, the mechanisms causing these variations seem unrelated since AO occurs much earlier in embryogenesis than the non-fusion of the ossification centres of the intramembranous portion of the occipital squama and wormian bones in the lambda region, which appear after the sixth fetal month [15]. It is interesting however, that *metopism* is frequently accompanied by supernumerary bones and remnants from other embryonic sutures [9, 21, 26, 35] as in this case. In view of the calvarial bone morphogenesis, these variations are closely interrelated and their

accumulation and overexpression are typical of some congenital disorders linked to impaired intramembranous bone formation.

In AO, a slight rotation of the midsagittal axis of the atlas, similar to that in the reported case, has been previously noted [2, 4, 5]. Unilateral paracondylar process, also called paramastoid process [12] is a common finding in AO [4, 12]. Paracondylar process could be connected to an epitransverse process of the transverse process of the atlas by a paracondylar-epitransverse articulation [11, 12]. In the here described case the right transverse process of the atlas is fused directly to the jugular process of the occipital bone, without formation of a paracondylar process (Fig. 3b). This additionally shortens the distance between the occipital bone and the atlas on the right side compared to the contralateral left side, where the transverse process is unfused. This results in an inclination of the atlas from the horizontal plane (Fig. 3a). According to Kadanoff and Mutafov [12], AO leads to formation of a new lower foramen magnum, called foramen occipitale (atlantooccipitale) magnum. It has been reported that AO is related to an abnormal shape of foramen magnum and a reduction of its dimensions, which may lead to neurological complications due to compression of the spinal cord and other nerves [2, 16]. In our case, the foramen magnum dimensions and the calculated area are slightly enlarged compared to the mean values (length 36.63 mm; breadth 31.47 mm; area 906.17 mm^2) reported for contemporary Bulgarian males [34], which is unusual. Similar to our case, it has been reported an irregular anterior atlantooccipital foramen formed between the superior aspect of the anterior arch of the atlas and the anterior margin of the foramen magnum with dimensions of 2–4 mm vertically and 18 mm horizontally [5]. Thus, the aperture is of similar height, but two times wider that this one in our case.

AO could impair the passage of either the vertebral arteries or C1 spinal nerves, which pass over the most anterior part of the posterior arch of the atlas. AO is frequently related to abnormalities in the vertebral artery and its route into the skull through accessory foramina and canals [2, 5, 16, 36]. If the posterior arch or hemiarch of the atlas is fused to the occipital bone, one should expect to find an anomalous osseous pathway for the vertebral artery to enter the cranium [36], canalis atlantooocipitalis a. vertebralis [12], as it is in the reported case. The abnormal passage may clamp on the arterial wall, thus narrowing the lumen of the vessel and decreasing the velocity of blood flow into the vertebral artery. The compression may result to ischemic symptoms, vertebro-basilar insufficiency and brainstem anoxia, while the artery

entrapment in a bony canal may cause dizziness, seizures, mental deterioration, syncope and rarely sudden death [16].

AO has also been linked to a reduction of the clivus length, inadequate or no formation of the atlantooccipital joint and flattening of the skull base [2, 16]. The measured *clivus* length in this case does not indicate such a reduction, and what is more interesting, the cranial base angle expresses an extensive flexion of the skull base instead of platybasia. It has been established that *metopism* is not related to a significant alteration in the cranial base angle [22], so the basilar kyphosis in this case is quite unusual.

Demineralization at the base of the dorsum sella is a valuable indicator of intracranial pathology. This appearance may be due to increased intracranial hypertension; it may also be secondary to a local mass lesion such as craniopharyngioma, optic chiasm glioma, or meningioma. An eroded posterior clinoid has also been reported with suprasellar aneurysm, histiocytosis X, and dilated third ventricle [30].

The interclinoid ligament joins the anterior and posterior clinoid processes while the caroticoclinoid ligament connects the anterior and middle clinoid processes. Sellar bridging has been recognized as an age-related phenomenon. Since clinoid bars have been commonly reported in fetuses and infants however, it seems more likely interclinoid bridging to reflect an abnormality at a much earlier stage in the *chondrocranium* development [7]. Interclinoid bridging is not pathological, but could cause many complications during surgical procedures in this area [3, 40]. Sellar bridging Type I results in the formation of anterior interclinoid foramen also known as caroticoclinoid foramen or arterial foramen through which the internal carotid artery passes. Bridging Type II gives rise to two separate foramina: caroticoclinoid foramen, and posterior interclinoid foramen, called as venous foramen which transmits the lateral part of circular sinus [3].

CONCLUSIONS

In the reported case, the observed variations in the *neurocranium* represent deviations from the normal developmental process arising at different stages during embryogenesis. The variations of the cranial base occur at an earlier developmental stage compared to that of the *calvaria*. The observed variations most commonly are asymptomatic and of little clinical significance, although they could be involved and/or cause serious health issues in some cases. In

the described case, there is no evident sign of a generalized disorder, which could cause the unusual co-occurrence of all this variations in the morphology of the *neurocranium*. Furthermore, bearing in mind that the individual was fit for military service, we can conclude that the man did not suffer from any severe complications.

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Figure 1. An adult male skull; a) parietal view; b) occipital view; c) basilar view.

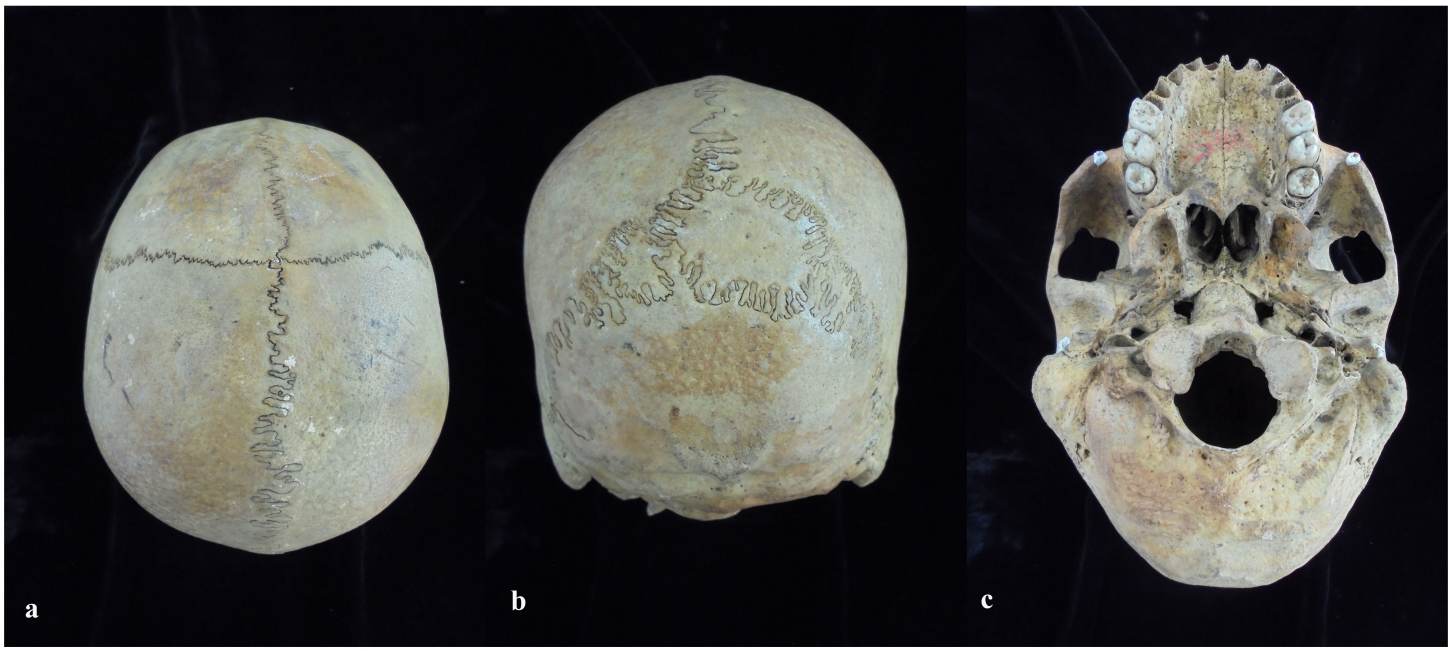
Figure 2. Volumetric rendering of the skull; a) frontal view: an entirely preserved metopic suture; b) occipital view: supernumerary calvarial bones at the interparietal portion of the occipital bone, a remnant from the mendosal suture (asterisk); c) right lateral view: an incomplete epipteric bone; d) basilar view: atlas occipitalization.

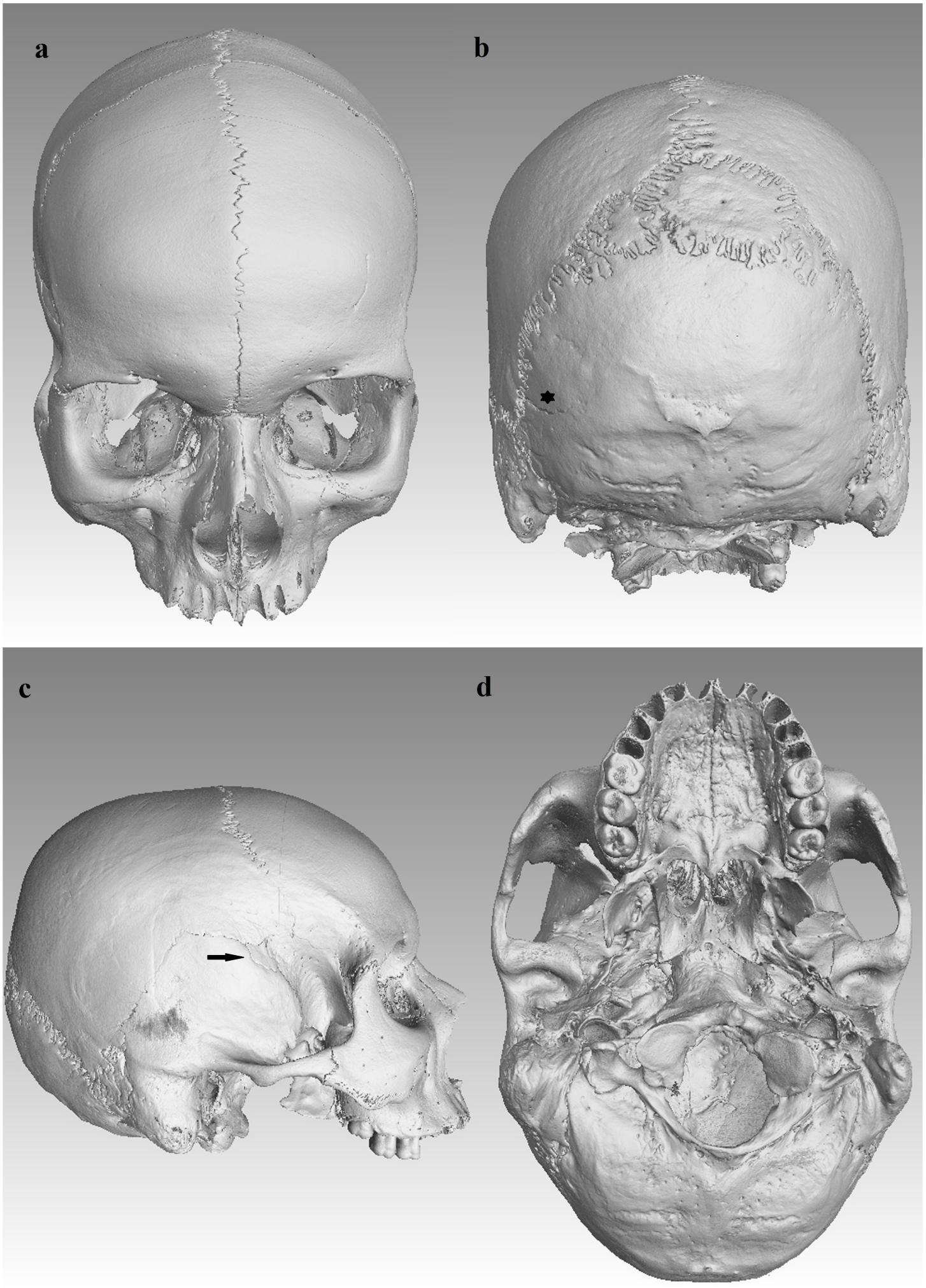
Figure 3. A close up view of the fused atlas; a) anterior view: an aperture between the fused foramen magnum margin and the anterior arch of the atlas (arrow); b) bilateral canals for the vertebral arteries passage (red arrows); a foramen formed between the fused transverse process of the atlas and the occipital bone (green arrow); a small vertical bony bridge between the occipital bone and the fused posterior arch of the atlas (black arrow).

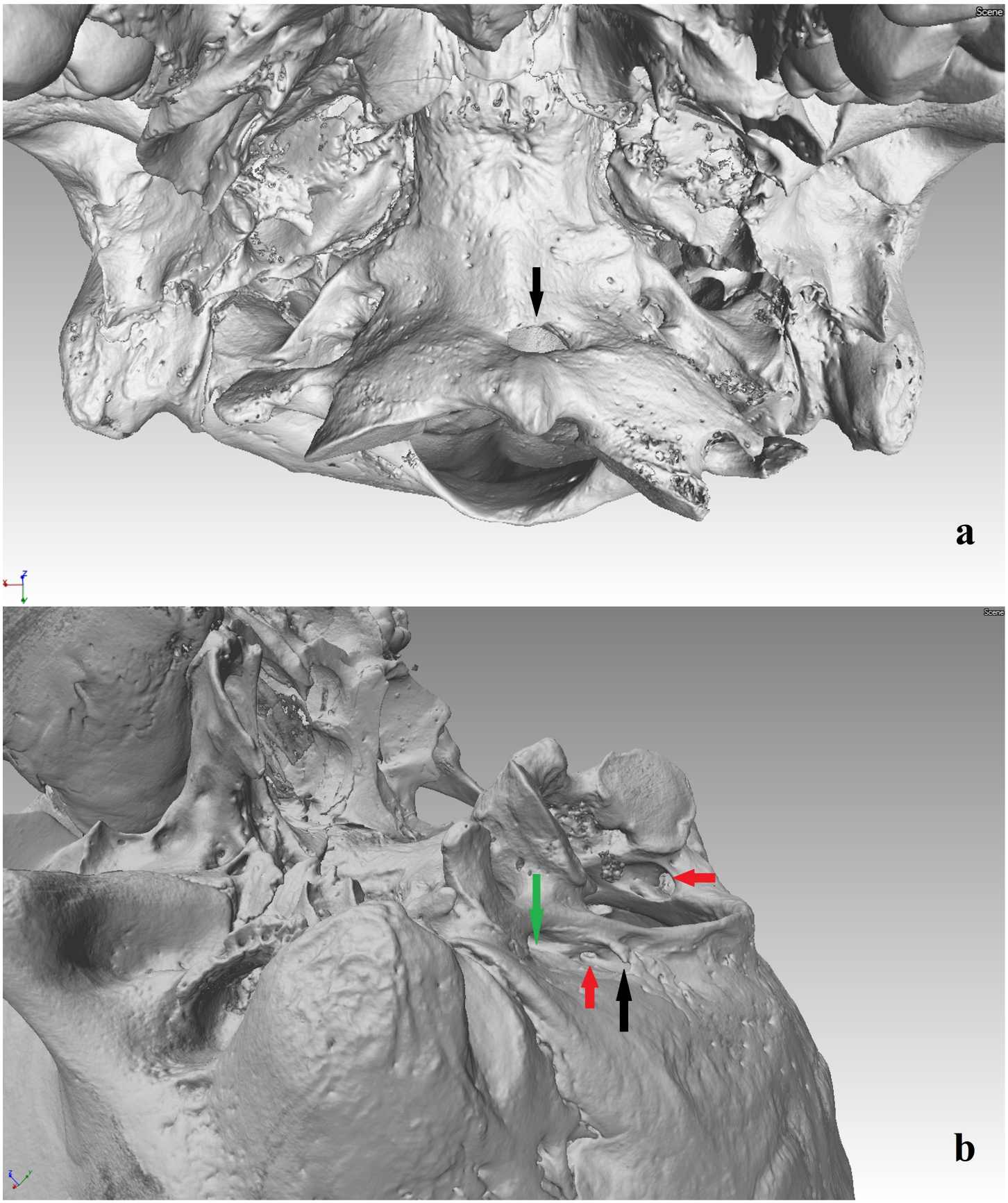
Figure 4. Middle cranial fossa: a) dorsum sellae; b) an interclinoid bridging Type II, mixed type (green arrow) on the right side enclosing an anterior interclinoid foramen (caroticoclinoid foramen) between the anterior and middle clinoid processes (black arrow), and a posterior interclinoid foramen between the middle and posterior clinoid processes (red arrow); c)

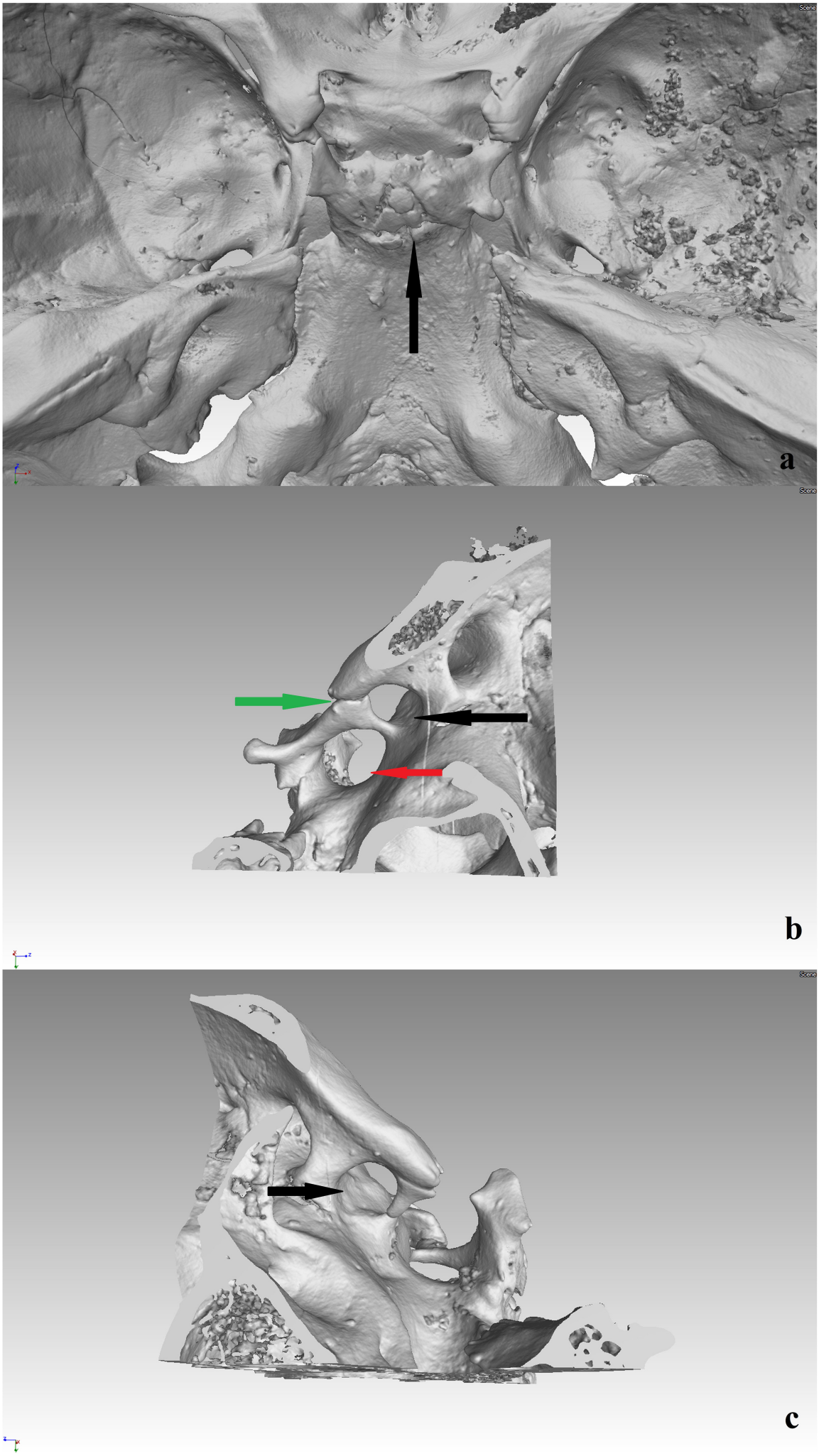
interclinoid bridging Type I, contact type representing a bone bridging between the anterior and middle clinoid processes, enclosing an anterior interclinoid foramen on the left side.

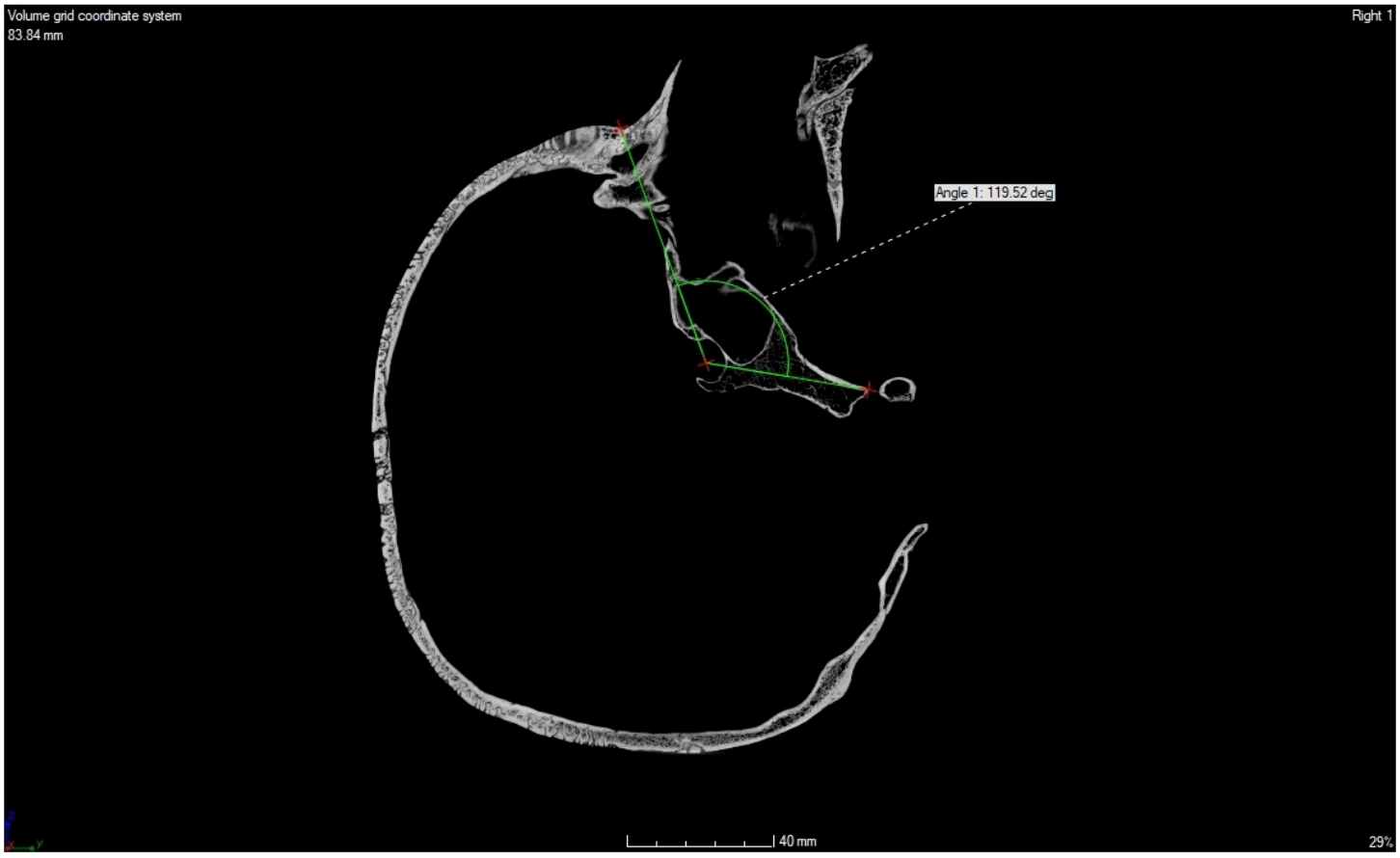
Figure 5. Cranial base angle constructed and measured in the midsagittal plane between the landmarks *nasion*, *sellae* and *basion*.


a
b
c


a
b
c
d


Scene
a
Scene
b


a
b
c


Volume grid coordinate system
83.84 mm
Right 1
Angle 1: 119.52 deg
40 mm
29%