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A case of early obliteration of the sagittal suture without effect on cranial deformation

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Abstract: This paper describes a unique case of craniosynostosis in a female skull in which sagittal sutures were completely fused by adolescence. Despite sagittal synostosis, the skull was of normal shape and size. Regarding craniometric features, the synostotic normocephalic skull was markedly different than that of scaphocephalic skulls which typically result from premature obliteration of the sagittal suture.

Keywords: sagittal suture, parietal bone, scaphocephaly, craniosynostosis.

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Introduction

The persistence of cranial sutures in early life allows for the growth of the brain. Premature closure of cranial sutures in early life may lead cranial deformations which are generally predictable. Early fusion of the sagittal suture, for example, limits brain growth laterally, thereby producing secondary frontal bossing and occipital bulging as the brain continues to grow [1]. The inverted "boat shape" of the cranium, resulting from the aforementioned sagittal synostosis, is typically referred to as scaphocephaly — a term used to differentiate the boat-shaped cranial morphology from other non-boat-shaped forms of dolichocephaly [2].

Sagittal synostosis is the most common form of craniosynostosis, accounting for 40–58% of all cases [3]. Isolated sagittal synostosis occurs in between 1 in 5,000 [4] to 1 in 4,200 children [5]. Of those afflicted with sagittal synostosis, approximately 78% are male [4, 6]. Sagittal synostosis is important to recognize because the resulting cranial deformation is, relative to non-craniostotic controls, more often accompanied by poorer language, learning, and memory [7], problems with vision or visual skills such as the ability to fix and follow [8–10], malocclusion [11], sleep-disordered breathing [12], and greater negative psychological impact regarding appearance [13, 14]. Furthermore, sagittal synostosis is often accompanied by elevated intracranial pressure [9, 15].

Though sagittal synostosis is associated with scaphocephaly, reports have noted isolated sagittal synostosis occurring without scaphocephaly [16]. Morritt *et al.* identified isolated sagittal synostosis without scaphocephaly in 8 of 193 (4.1%) isolated sagittal synostosis cases [15]. All such cases were among children under the age of seven years. Further, seven of the eight cases, had remarkable cranial findings including turricephaly, brachycephaly, plagiocephaly, central forehead bulging, metopic ridge, vertex saddle, microcephaly, and macrocephaly. One case, however, was a normocephalic female of five years and nine months age. In six of the eight patients, intracranial pressure was measured and, subsequently, revealed high intracranial pressure in four (67%), including the aforementioned normocephalic female [15].

A study of sagittal synostosis among 3,636 adult crania (≥20 year of age), revealed no relationship between age and the extent of suture closure [17]. Indeed, the study noted the frequency of premature closure in the 3rd–7th decades was 2.0%, 3.4%, 2.0%, 2.5%, 2.7%, respectively. Among crania of individuals older than 70 years, the frequency of suture closure was 0.7% [17]. Extremely rare isolated craniosynostosis occurring among normocephalic children under the age of six years has been described [15]. Also, sagittal synostosis has also been described in adults over the age of 20 years [17]. Our study documents a normocephalic cranium from an individual of late adolescence with isolated sagittal synostosis.

Case Report

Inspection of the museum collection of the Department of Anatomy of the Medical College of the Jagiellonian University revealed a dry human skull with evidence of isolated complete absence of the sagittal suture. Neither the ethnicity, date of origin, nor any other demographic information was available. A brief note included in the inventory book dated from 1874 states that the skull was that of a 20-year-old individual (Fig. 1). Indeed, the degree of cranial suture closure and non-ossified spheno-occipital cartilage suggests that this skull may derive from an individual being in adolescent age at death.

The skull morphology indicates the female sex: vertically-oriented forehead, non-pronounced glabella and supraorbital rims, rounded orbits, subtle zygomatic bones, a delicate mandible with non-pronounced gonions. The mastoid processes are small. Likewise, the external occipital protuberance and nuchal crests are slightly developed (Fig. 2).

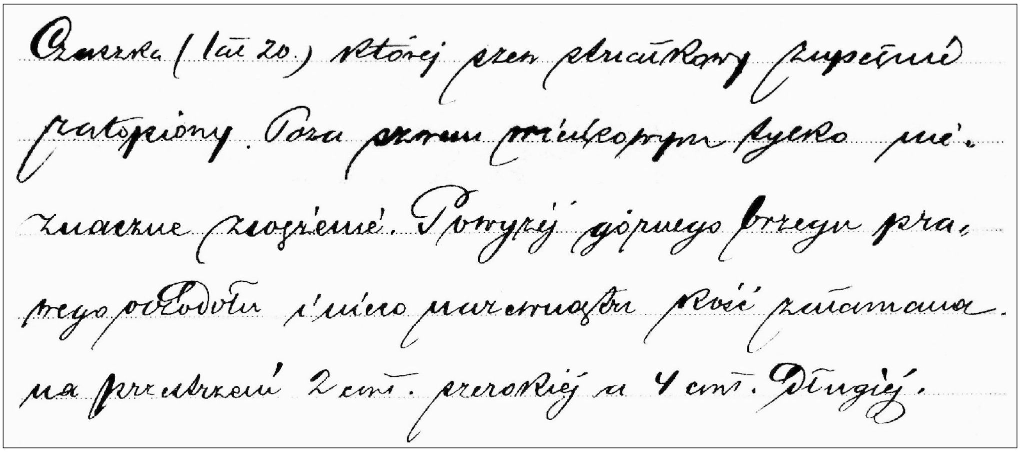
Czaszka (lat 20.) której szew strzałkowy zupełnie
zarośnięty. Poza szwem wieńcowym tylko nie-
znacznie zrośnięte. Powyżej górnego brzegu pra-
wego oczodołu w miejscu [illegible] kość [illegible]
na przestrzeni 2 cm. szerokiéj a 4 cm. długiéj.

Fig. 1. A photocopy showing a brief hand-written description (in the Polish language) of the investigated skull taken from the 19th century inventory book of the Anatomical Museum of the Department of Anatomy of the Medical College of the Jagiellonian University.

Neither frontal bossing nor occipital prominence were present. Likewise, there was no so-called "saddle defect," which is typical of sagittal synostosis [18]. Also, there was no persistence of the metopic suture, which has been postulated to allow for normal anterior cranial growth in the setting of isolated sagittal synostosis [15]. On the ectocranial surface of the calvaria, there were perceptible traces resembling obliterated sutures, lateral to the median sagittal plane. The outlines of both presumable sutures reach neither the coronal nor lambdoid sutures. Applied investigation techniques did not explain whether the laterally positioned scratch-like bony surface features are remnants of atypical sutures (Fig. 3).

The skull had an ovoid vault. The calvaria was relatively short and narrow with a steeply rising frontal bone and moderately inclined forehead. The frontal squama in the sagittal profile presented a distinct arc. Posteriorly, the cranial roof was strongly curved, descending to the occipital squama. The occipital plane was much longer than the nuchal plane and nearly perpendicular to the cranial base. The occiput was moderately protuberant posteriorly. The lambdoid suture was weakly interdigitated, not obliterated, and was interrupted by a single Wormian bone on the left side.

The temporal lines were well expressed on both sides of calvaria and the squama of the temporal bone was large on both sides of the skull. The bony face was relatively narrow. The alveolar prognathism and bilaterally well-marked maxillary incisure were characteristic for this skull. The right orbit was larger than the left. The nasal bones were wavy-convex.

Cephalometric measurements of the cranium were acquired with instruments including sliding callipers, spreading callipers, and flexible tape, according to estab-

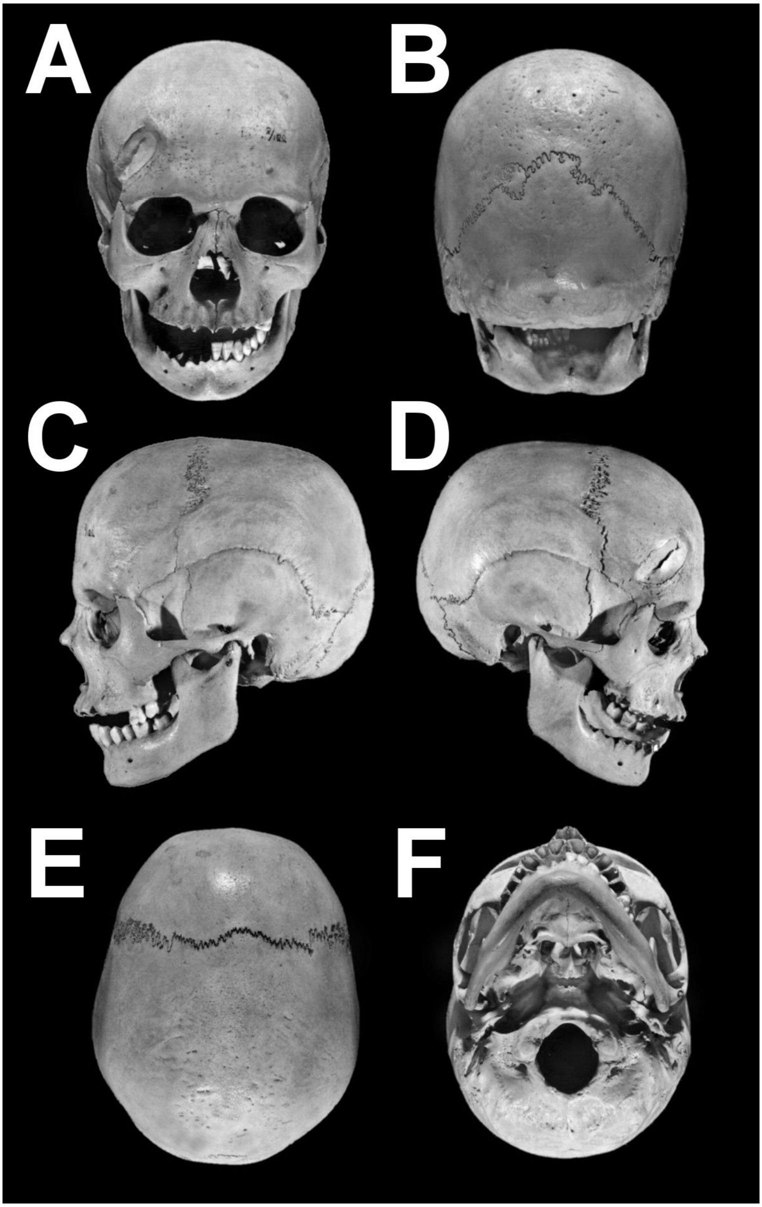

Fig. 2. Normocephalic skull with isolated sagittal synostosis. **A:** Norma frontalis view demonstrating absence of metopism. Likewise, there is no pronounced ridging in the midline. **B:** Norma occipitalis view demonstrating a sutural bone within the left side of the lambdoidal suture. **C:** Left norma lateralis view demonstrating a non-scaphocephalic shape of the cranial vault. **D:** Right norma lateralis view demonstrating normal cranial shape. **E:** Norma verticalis view demonstrating absence of the sagittal suture, absence of the metopic suture, and an open coronal suture. **F:** Norma basalis view with evidence of a large gap consistent with a non-ossified spheno-occipital synchondrosis.

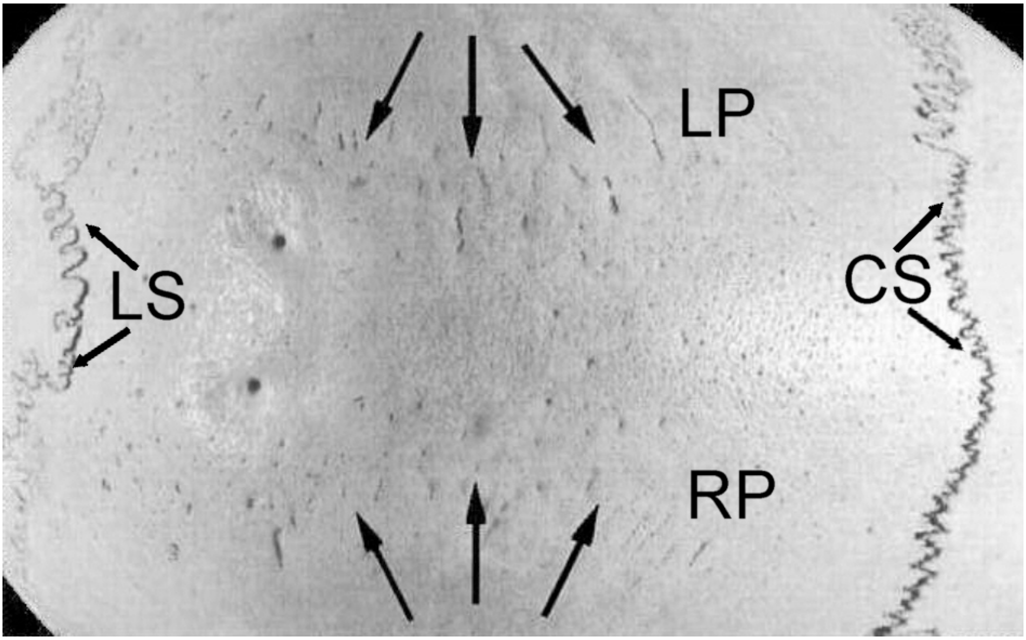

Fig. 3. Remnants of probable uneven sutures (indicated by the arrows) that might have existed bilaterally on the cranial vault (parietal area); LP — left parietal bone, RP — right parietal bone, LS — lambdoid suture, CS — coronal suture.

lished methodology defined in Table 1 [19, 20]. The same measurements were also taken of a scaphocephalic skull, also housed in the Department of Anatomy of the Medical College of the Jagiellonian University, for the purpose of comparison.

Table 1. Definitions of the craniometric measurements performed in this study.

Parameter	Definition
Maximum cranial length (g-op)	Distance between glabella and opisthocranion in the midsagittal plane, measured in a straight line
Maximum cranial breadth (eu-eu)	Maximum width of skull perpendicular to mid-sagittal plane wherever it is located, with the exception of the inferior temporal lines and the area immediately surrounding them
Bizygomatic diameter (zy-zy)	Direct distance between most lateral points on the zygomatic arches
Basion-bregma height (ba-b)*	Direct distance from the lowest point on the anterior margin of foramen magnum to bregma
Cranial base length (ba-n)	Direct distance from nasion to basion
Basion-prosthion length (ba-pr)	Direct distance from basion to prosthion
Nasion-prosthion length (n-pr)	Direct distance from nasion to prosthion

Table 1. cont.

Parameter	Definition
Nasion-gnathion length (n-gn)	Direct distance from nasion to gnathion
Minimum frontal breadth (ft-ft)	Direct distance between the two frontotemporale
Frontal chord (n-b)*	Direct distance from nasion to bregma taken in the midsagittal plane
Parietal chord (b-l)*	Direct distance from bregma to lambda taken in the midsagittal plane
Occipital chord (l-o)*	Direct distance from lambda to opisthion taken in the midsagittal plane
Arc length (n-b)*	Arch length measured along the squama of the frontal bone between nasion and bregma in the midsagittal plane
Arc length (b-l)*	Arch length measured between bregma and lambda along the parietal bone in the mid-sagittal plane
Arc length (l-o)*	Arch length measured between lambda and opisthion along the squama of the occipital bone in the mid-sagittal plane

* Because of the absence of the sagittal suture in the investigated skull, the bregma and lambda landmarks were demarcated arbitrarily in the place of intersection of the coronal and lambdoid sutures with the median sagittal plane, respectively.

Detailed reports about the scaphocephalic skull have been published previously [21, 22]. In order to compare the dimensions of the skull having the obliterated sagittal suture with normal skulls, a control dataset of Polish male and female skulls was taken from the literature [23]. It should be stressed that the values of craniometric measures can serve only as the exemplary data, typical for the selected population regarded as the normocephalic subjects. Due to the unknown geographical and historical origin of the skull with the lack of the sagittal suture, craniometrical comparison has limited value and depicts only potential differences between various forms of human skulls. Comparison of the craniometric features provided further confirmation of the normocephalic structure of the skull and its deviation from scaphocephaly (Table 2).

The skull with obliterated sagittal suture was also subjected to radiographic examination in order to visualize the inner structure of the cranial bones in the region of the sagittal suture. For this purpose we applied a standard radiological protocol dedicated for bone imaging which was performed with the aid of the Siemens Somatom Sensation CT scanner. Further, three-dimensional renderings were extracted from CT scans to assess the inner and outer surfaces of the cranial vault. The texture of the bone where the sagittal suture normally exists was completely homogenous in nature and did not show any traces of an earlier existence of the suture (Fig. 4).

Table 2. Craniometric features (in millimeters) of the skull with obliterated sagittal suture versus scaphosephalic skull and normocephalic control group.

Measurement	Scapho-cephalic skull	Skull with absent sagittal suture	Control group Mean values					
			Female	±SD	N	Male	±SD	N
g-op	201	175	171.0	6.49	87	179.4	7.31	83
ba-b	130	134	128.2	5.02	70	134.6	6.77	60
eu-eu	127	137	142.3	4.53	87	148.6	5.45	80
n-b	120	111	106.8	4.76	81	112.5	3.48	75
b-l	144	115	106.0	5.73	87	111.6	5.23	83
l-o	85	93	90.9	5.92	73	94.7	5.40	76
ft-ft	103	93	97.0	3.82	82	98.7	5.15	77
n-gn	113	112	111.2	6.56	11	112.1	4.60	10
ba-n	92	98	94.7	4.26	67	100.1	6.17	54
ba-pr	84	94	91.0	4.10	40	95.6	6.70	34
n-pr	60	61	63.8	4.83	43	68.6	3.86	42
zy-zy	130	123	126.1	4.38	33	134.0	5.16	31
arc n-b	147	131	122.3	6.27	81	126.2	5.22	76
arc b-l	160	129	118.8	7.40	86	124.6	9.64	83
arc l-o	110	110	111.2	7.41	73	115,5	7.07	74

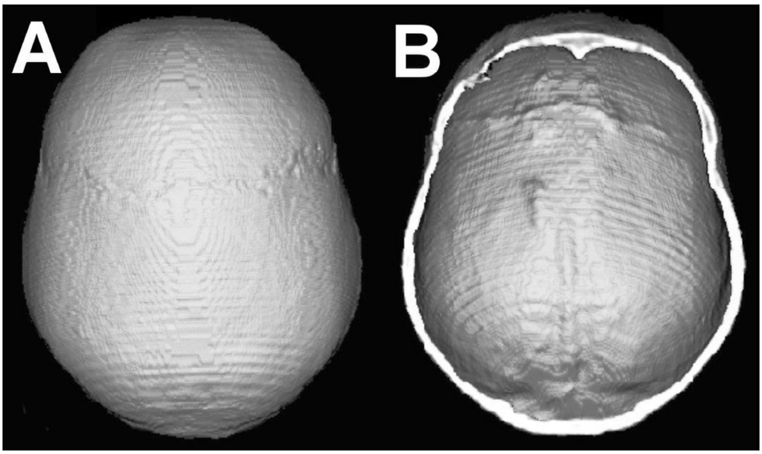

Fig. 4. Computed tomography volume reconstruction of the calvaria. **A:** Ectocranial view demonstrating absence of the sagittal suture. **B:** Endocranial view demonstrating absence of the sagittal suture. The groove for the superior sagittal suture located just right of the midline. The right side of the parietal bone, anterior aspect, and frontal bone have evidence of mild thumb-printing which may be indicative of elevated intracranial pressure.

Discussion

Cranial sutures play a crucial role in the proper growth of the skull. Premature closure of the cranial sutures causes overall or local deformation of the skull resulting in altered shape of the cranial vault. [24, 25]. Beside common forms of scaphocephaly, other entities of the craniosynostosis involving sagittal suture have been reported in children, for example: 'Mercedes Benz' pattern of synostosis (bilateral lambdoid and sagittal synostosis), 'Z-pattern' synostosis (left coronal, sagittal, and right lambdoid) or synostosis of the sagittal suture which extended on the squama of the occipital bone. Each of the aforementioned craniosynostoses are known to result in an altered shape of the skull [26–28].

As it was mention in the Introduction section, premature obliteration of the sagittal suture usually gives a keel-like shape to the skull. Such a skull is described as having a midline bony ridge over the sagittal suture region, biparietal and bitemporal narrowing, and occipital prominence. These morphological features were not observed in our case, and the shape of the cranial vault was definitely unlike the scaphocephalus, despite that both parietal bones were fused before the age of 20. Hence, its relatively early obliteration did not disturb proper growth the skull, thus the head attained normal shape and size. The period when the sagittal suture obliterates is quite variable and usually begins between 30 and 40 years of age and progresses for the subsequent 10 years. Alves *et al.* found that the sagittal suture has been entirely preserved between 20–35 years [29]. However, early obliteration of this suture does not have to be associated with a pathological condition such as craniosynostosis; rather, it may be a case due to atypical timing of suture patency. Descriptions of non-deformed skulls without the sagittal suture are rare in the scientific literature and limited to only a few osteological findings. Thereby, this case highlights extreme variability in the time of closing of the sagittal suture due to its physiological ossification, which remains in contrast to pathological ossification leading to the scaphocephaly. Occurrence of such synostosis, like that in this case, does not alter the normal shape of the head, thereby individuals with a fused sagittal suture and a normally shaped skull do not require cranial surgery in contrast to pathological craniosynostosis which induces skull and brain deformations.

Conclusions

In spite of the apparent absence of the sagittal suture, being an effect of its early obliteration, this report demonstrates substantial morphometric differences between this skull and a scaphocephalic skull, but not between this skull and normocephalic skulls. Therefore, the examined skull appears a rare case of early sagittal synostosis that resulted in an unremarkable effect on the cranial shape.

Conflict of interest

None of the authors have any conflict of interest nor any financial interest.

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