### ORIGINAL ARTICLE

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# How does occipitalization influence the dimensions of the cranium?

#### **Structured Abstract**

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**Objectives** – To describe occipitalization on human dry skulls and to compare craniofacial morphology including the posterior cranial fossa in skulls with occipitalization and in skulls without occipitalization and with normal craniofacial morphology (controls).

**Setting and Sample Population** – A total of 29 skulls were selected from the Björk collection. Nine had occipitalization of atlas. Twenty had no dentoalveolar or craniofacial anomalies, and no occipitalization was visible on the skulls (controls). The ages of the skulls were between adolescence and adulthood.

*Methods* – Visual assessments to describe the occipitalization pattern, direct measurements to measure the foramen magnum dimensions and cephalometric radiographic analyses to describe the craniofacial morphology. New variables were determined to describe the posterior cranial fossa.

**Results** – Of the skulls with occipitalization, 67% had complete and 33% had partial occipitalization. In the posterior part of the neural arch, 44% had a cleft. Occipitalization was significantly associated with a small foramen magnum (p < 0.01; p < 0.001) and deviant shape of the posterior cranial fossa (p < 0.05). The craniofacial morphology in the vertical and sagittal dimensions including the cranial base angle was normal in skulls with occipitalization, which indicates that occipitalization is associated with deviant morphology of the surrounding bony structures but not with deviations in the craniofacial morphology in general. The results of this study are important for the diagnosis of occipitalization on 2D radiographs.

**Key words:** craniofacial dimensions; dry skulls; foramen magnum; occipitalization; posterior cranial fossa

# Introduction

Occipitalization is defined as assimilation of the atlas with the occipital bone (1) and can be referred to as occipito-atlas synostosis (2). Occipitalization can be partial or complete (3–7) or unilateral or bilateral (4, 5, 7, 8). In cases with partial occipitalization, the fusion is located in the anterior arch, the posterior arch, the lateral masses of the atlas or in two or more of these locations (3, 7, 9). Most common is partial occipitalization (7, 10). Cleft of the atlas arches can also occur in combination with occipitalization. In these cases, the cleft is most commonly located in the

posterior arch (11). Occipitalization occurs in 0.08– 2.76% of the population (12–16), men and women equally affected (5, 12, 16–19). Occipitalization can occur as a developmental defect in somite segmentation or as a failure in the re-segmentation between the fourth occipital sclerotome and the first cervical vertebra in early embryogenesis (11, 16, 20–22). Occipitalization of the atlas occurs in syndromes such as Klippel-Feil (1, 7), Down and Goldenhar (1, 23–25).

Cephalometric studies on profile radiographs have shown that the horizontal and vertical dimensions of the atlas are associated with cranial base angulations and with mandibular shape and growth (26–28). Other cephalometric studies have shown that morphological deviations of the cervical vertebral column, such as fusion anomalies including occipitalization, are associated with a large cranial base angle, retrognathia of the jaws and inclination of the jaws (29–34). These studies suggest an association between cervical vertebral column morphology including occipitalization and craniofacial morphology.

From an embryological point of view, the extension of the notochord in the early body axis from the spine to the sella turcica (35) may explain the developmental association between the vertebral column and the posterior part of the occipital bone. This association has recently been illustrated on postnatal profile radiographs (36).

Two studies of dry skulls with occipitalization of the atlas found irregular margins and shapes of the foramen magnum (37, 38). One of these studies revealed that the length of the clivus was reduced, whereas the cranial base angle was normal (37). Both studies suggest an association between occipitalization and the dimensions of the foramen magnum.

The aims of this study were 1) to describe patterns of occipitalization on human dry skulls; 2) to compare the craniofacial morphology including the posterior cranial fossa in skulls with occipitalization; and 3) to compare the craniofacial morphology including the posterior cranial fossa in skulls with occipitalization with that of skulls without occipitalization and with normal craniofacial morphology (controls).

# Material and methods

From the Björk collection of 223 normal and pathological human dry skulls (the Department of Orthodontics, Copenhagen School of Dentistry, Denmark), 29 skulls were selected for this study. Nine of these 29 skulls had occipitalization of the atlas. Twenty had no dentoalveolar or craniofacial anomalies, and no occipitalization was visible on the skulls (controls). The ages of the skulls were between adolescence and adulthood.

Visual and direct measurements on the skulls and cephalometric analyses on profile radiographs were performed.

#### Visual assessment

#### Occipitalization patterns

Occipitalization was defined as an osseous continuity of the occiput and atlas as described by Smoker (8) and was divided into three zones according to Gholve et al. (7).

- Zone 1 (Z1): occipitalization of the anterior arch of the atlas.
- Zone 2 (Z2): occipitalization of the lateral masses of the atlas.
- Zone 3 (Z3): occipitalization of the posterior arch of the atlas (Fig. 1).

#### Clefts

The skulls with occipitalization were divided into two groups according to occurrence of clefts: One group with cleft of the posterior portion of the neural arch of



*Fig. 1.* Caudal view of occipitalization of the atlas. The thin lines divide the anatomical parts of the atlas as defined by Gholve et al. (7): the anterior arch zone (Z1), the lateral masses zone (Z2) and the posterior arch zone (Z3). The thick lines illustrate the transversal and sagittal dimension measured directly on the skull according to Al-Motabagani & Surendra (37).

the atlas and one group without cleft. The cleft was defined according to Von Torklus (16) as a cartilaginous cleft, an open synchondrosis, between the osseous posterior neural arches of the atlas.

#### Direct measurements

The dimensions of the foramen magnum were measured by the length of the sagittal and transversal dimensions (Fig. 1). The area of the foramen magnum was calculated directly on the skull using a formula according to Al-Motabagani & Surendra (37):  $\pi$ ·0.25*·sagittal·transversal*.

#### Cephalometric analyses

A profile radiograph was taken of each skull, placed in the Frankfort horizontal plane, at the Department of Radiology, School of Dentistry, University of Copenhagen, Denmark, in a Philips/Valmet BR 2002 cephalostat with a film focus distance of 195 cm. The linear enlargement was 8.3%. The radiographic film used was LifeRay XDA Plus UTLG (Ferrania Technologies S.p.A., Cairo Montenotte, Italy). The films were exposed with 65-67 kv and 5-7 mA.

The cephalometric analyses were performed to describe the craniofacial morphology and the posterior cranial fossa. The variables describing the craniofacial morphology were defined according to Björk (39). In one skull, the mandible was missing and therefore the variables s-n-pg, NSL/ML, n-pg and gn-tgo-ar were measured in only eight of the nine skulls with occipitalization.

New measurement variables were defined to describe the posterior cranial fossa. The new variables are:

- s-d: the length from the sella point(s) to the deepest point in posterior cranial fossa.
- s-iop: the length from the sella point(s) to the internal occipital protuberance.
- d-p: the length from the deepest point in the posterior cranial fossa (d) perpendicular to s-iop (p).
- p-iop: the length from the internal occipital protuberance (iop) to the point p.
- iop-s-d: the angle between the s-d and the s-iop lines.
- Tt: Theca thickness is the thickness of the cranium measured at the internal occipital protuberance.

The variables are shown in Table 1 and Fig. 2.

The locations of all reference points and lines were checked by one of the co-authors (LS). The method error ranged from 0.09 to 0.69 degrees (40) and the reliability coefficients from 0.99 to 1.00 (41).

#### Statistical analysis

The normality of the distribution was assessed by parameters of skewness and kurtosis and by the Shapiro-Wilks *W*-test. Measurements of the craniofacial morphology, the posterior cranial fossa and the foramen magnum dimensions were normally distributed except for Ba-s-theca and n-pg, which deviated slightly from the normal distribution. Differences in means of the craniofacial morphology, the posterior cranial fossa and foramen magnum dimensions between the groups of skulls with occipitalization and the controls were assessed by unpaired *t*-test.

#### Results

#### Visual assessment

Occipitalization patterns

In six skulls (67%), the occipitalization occurred in zones 1, 2 and 3 (complete occipitalization) (Fig. 3). In three skulls (33%), the occipitalization occurred in zones 2 and 3 (partial occipitalization) (Fig. 4).

#### Clefts

In four skulls with occipitalization (44%), a cleft in the posterior part of the neural arch occurred. No cleft was seen in the remaining five skulls with occipitalization (56%) (Figs 3 and 4).

#### Associations

#### Craniofacial morphology

Regarding craniofacial morphology in the vertical and sagittal dimensions, no significant differences were found within the occipitalization groups or between the skulls with occipitalization and the controls (Table 1).

#### The posterior cranial fossa

The distance between the sella turcica and the deepest point in the posterior cranial fossa (s-d) was significantly larger (p < 0.05) in the cleft group when compared to the non-cleft group. The distance between the internal occipital protuberance (iop) to the point (p):the line from the deepest point in posterior cranial

Variable (degrees)	Control $(n = 20)$		Occipit	alizatio	on												
			( <i>n</i> = 9)		Complete $(n = 6)$			Partial $(n = 3)$			Cleft $(n = 4)$			No cleft $(n = 5)$			
	Mean	SD	Mean	SD	р	Mean	SD	р	Mean	SD	р	Mean	SD	р	Mean	SD	р
Measurements on p	orofile ra	diogra	phs														
Craniofacial morphol	ogy																
Sagittal dimensions																	
s-n-ss	85.1	3.4	84.8	5.5	NS	86.3	6.3	NS	81.7	1.3	NS	86.5	5.0	NS	83.4	6.0	NS
s-n-pg	80.9	3.5	80.8	6.4	NS	80.6	7.1	NS	81.5	5.7	NS	81.5	5.7	NS	83.3	3.8	NS
Vertical dimensions																	
NSL/NL	9.5	3.9	10.9	3.3	NS	10.9	4.1	NS	10.8	1.0	NS	9.9	0.6	NS	11.7	4.4	NS
NSL/ML	32.1	5.8	34.4	5.2	NS	34.5	5.3	NS	34.0	7.1	NS	33.9	4.5	NS	34.9	6.5	NS
n-ss (mm)	52.7	5.3	53.2	3.3	NS	53.0	4.1	NS	53.7	1.5	NS	51.3	3.0	NS	54.8	30.0	NS
n-pg (mm)	105.2	8.7	105.3	5.9	NS	103.2	5.3	NS	111.5	0.7	NS	105.8	8.3	NS	104.8	3.7	NS
Gn-tgo-ar	123.5	8.7	124.7	4.7	NS	124.3	4.8	NS	125.8	6.0	NS	127.4	4.9	NS	122.0	2.9	NS
Cranial base																	
N-S-Ba	131.6	5.1	134.8	4.8	NS	135.2	5.4	NS	134.0	4.4	NS	134.9	6.2	NS	134.7	4.3	NS
n-s (mm)	65.7	4.2	67.8	4.0	NS	67.7	4.3	NS	68.0	4.0	NS	68.3	3.9	NS	67.4	4.5	NS
s-ba (mm)	39.5	3.7	39.2	4.9	NS	37.7	4.1	NS	43.0	4.6	NS	40.3	6.0	NS	38.4	4.3	NS
Posterior cranial foss	a																
s-d (mm)	66.1	4.4	64.2	7.4	NS	61.8	7.4	NS	69.0	5.6	NS	69.5	3.7	*	60.0	7.0	*
p-iop (mm)	29.7	3.4	30.4	5.2	NS	31.3	6.0	NS	28.7	2.9	NS	26.8	4.5	*	33.4	3.6	*
s-iop (mm)	89.1	3.5	89.7	3.9	NS	88.2	3.7	NS	92.7	2.5	NS	90.0	5.3	NS	89.4	3.1	NS
d-p (mm)	29.0	3.1	30.9	3.8	NS	32.5	3.5	NS	27.7	1.5	NS	29.5	1.7	NS	32.0	4.7	NS
iop-s-d (mm)	48.1	3.1	46.6	6.2	NS	46.3	7.8	NS	47.0	1.7	NS	42.3	7.0	NS	50.0	2.6	NS
tt (mm)	13.4	3.3	12.9	2.6	NS	12.8	2.8	NS	13.0	2.6	NS	13.5	1.7	NS	12.4	3.2	NS
Measurements dire	ctly on th	ne sku	lls														
Foramen magnum di	mension	6															
Sagittal (mm)	33.5	1.9	30.2	3.8	**	28.5	2.9	***	33.7	3.2	NS	32.5	3.5	NS	28.4	3.2	**
Transversal (mm)	28.3	2.3	26.4	3.6	NS	25.0	3.4	NS	29.1	2.3	NS	27.1	3.2	NS	25.8	4.2	NS
Area (cm <sup>3</sup> )	7.5	0.9	6.3	1.4	**	5.6	0.7	****	7.7	1.4	NS	6.9	1.5	NS	5.8	1.2	**

# Table 1. Associations between occipitalization and the craniofacial morphology including the posterior cranial fossa and foramen magnum dimensions

\* $p \le 0.05$ , t-test within the individual groups.

\*\* $p \le 0.05$ , t-test between controls and the individual groups.

\*\*\* $p \le 0.01$ , t-test between controls and the individual groups.

\*\*\*\* $p \le 0.001$ , t-test between controls and the individual groups.

fossa perpendicular to the line between sella turcica and the internal occipital protuberance (p-iop) was significantly smaller (p < 0.05) in the cleft group compared to the non-cleft group (Table 1, Fig. 2).

#### The foramen magnum

The sagittal dimensions and area of the foramen magnum were significantly smaller in skulls with occipitalization (p < 0.05), significantly smaller in skulls with complete occipitalization (p < 0.01; 0,001)

and significantly smaller in skulls without cleft in the posterior arch (p < 0.05) when compared to controls (Table 1).

## Discussion

In this study, the occipitalization patterns of human dry skulls were described and associations between occipitalization and dimensions of the cranium were found.



*Fig. 2.* Schematic drawing of the cephalometric measurements describing the craniofacial morphology including the posterior cranial fossa. The variables describing the craniofacial morphology were defined according to Björk (39). The variables describing the posterior cranial fossa were defined as: s-d: the length from the sella turcica to the deepest point in posterior cranial fossa; s-iop: the length from the sella turcica to the internal occipital protuberance; d-p: the length from the deepest point in the posterior cranial fossa (d) perpendicular to s-iop (p); p-iop: the length from the internal occipital protuberance (iop) to the point p; iop-s-d: the angle between the s-d and the s-iop lines; Tt: Theca thickness is the thickness of the cranium measured at the internal occipital protuberance.

#### **Occipitalization patterns**

In 6 out of 9 skulls with occipitalization, complete occipitalization occurred and in 3 out of 9, partial occipitalization occurred. This is in disagreement with previous studies reporting that partial occipitalization is most common (7, 10). In this study, cleft of the arches of atlas was only seen in the posterior arch of atlas. This is in agreement with findings from a previous study concluding that cleft is most commonly observed in the posterior arch of atlas (11).

#### Associations

In this study, a new association was observed between occipitalization and the posterior cranial fossa. In the cleft group compared to the non-cleft group, the distance between the sella turcica and the deepest point in



*Fig. 3.* Occipitalization of the atlas with fusion in zones 1, 2 and 3. No cleft of the posterior arches. (A) caudal ventral view. White arrow shows no cleft of the posterior portion of the posterior arch. (B) caudal dorsal view. Fusions are marked by black stars. Note that the foramen magnum is small and round and narrowed by the lateral masses.

the posterior cranial fossa was significantly larger and the distance between the internal occipital protuberance to the line from the deepest point in the posterior cranial fossa perpendicular to the line between the sella turcica and the internal occipital protuberance was significantly smaller. These findings indicate that the shape of the posterior cranial fossa is significantly associated with occipitalization with and without cleft of the posterior arch of atlas.

Furthermore, an association between occipitalization and dimensions of the foramen magnum was found. The sagittal dimensions and the area of the foramen magnum evaluated on the skulls were significantly smaller in skulls with occipitalization compared to controls. These findings indicate that occipitalization is significantly associated with a small foramen magnum. Previous studies have also found an association between occipitalization and the foramen magnum (37, 38). These studies found irregular margins and shapes of the foramen magnum in dry skulls with occipitalization.

As occipitalization may be missed on 2D radiographs, the association found in this study between occipitalization, size of the foramen magnum and shape of the posterior cranial fossa may help to determine whether occipitalization occurs in a patient. If occipitalization does occur, a small foramen magnum and a deviant shape of the posterior cranial fossa should be observed radiographically. Even though 3D imaging would identify occipitalization, most clinics



*Fig. 4.* Occipitalization of the atlas with fusion in zones 2 and 3. No cleft of the posterior arches. (A) caudal ventral view. White arrow shows no cleft of the posterior portion of the posterior arch. (B) caudal dorsal view. Fusion is marked by a black star. Note that the foramen magnum is small and round and narrowed by the lateral masses.

currently only have 2D imaging. Radiographic material used until recently is 2D only. For evaluation of these 2D radiographs, this study is considered valuable. Furthermore, the results of this study serve as a diagnostic tool to help diagnose occipitalization on 2D radiographs still in use in daily clinical practice.

Recent cephalometric studies have shown that morphological deviations of the cervical vertebral column such as fusion anomalies were associated with a large cranial base angle, retrognathia of the jaws and inclination of the jaws (29–34). An association between occipitalization and craniofacial morphology in the vertical and sagittal dimensions was expected but was not found in this study. In agreement with a previous study (37), the cranial base angle was normal in skulls with occipitalization. This indicates that occipitalization is associated with the surrounding bony structures and not with the craniofacial morphology in general.

In this study, new cephalometric variables were defined to describe the posterior cranial fossa on profile radiographs of human dry skulls with occipitalization and to further elucidate the association between occipitalization and craniofacial morphology. Deviations in the posterior cranial fossa observed on profile radiographs could be a sign of occipitalization. Therefore, specific attention to this area may help to make a precise diagnosis of occipitalization.

# Conclusions

This study concludes that occipitalization is significantly associated with a small foramen magnum and deviant shape of the posterior cranial fossa. It also concludes that the craniofacial morphology in the vertical and sagittal dimensions including the cranial base is normal in skulls with occipitalization. This indicates that occipitalization is associated with the surrounding bone structures and not with deviations in the craniofacial morphology. The results of this study serve as a diagnostic tool to help diagnose occipitalization on 2D radiographs in daily clinical practice.

# Clinical relevance

This study found an association between occipitalization and a small foramen magnum and a new association between occipitalization and a deviant shape of the posterior cranial fossa. As occipitalization is difficult to visualize on 2D radiographs, this association may help to determine whether occipitalization occurs in a patient. If so, a small foramen magnum and deviant shape of the posterior cranial fossa should be observed radiographically. The results of this study serve as a diagnostic tool to help diagnose occipitalization on 2D radiographs in daily clinical practice.

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