

THE INCIDENCE OF ATLANTO-OCCIPITALIZATION AND ADDITIONAL FORAMINA PRESENT IN THE DRY SKULLS OF NEPALESE POPULATION

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ABSTRACT

Introduction

Atlanto-occipitalization (AOZ) is one of the congenital anomalies related to craniovertebralsynostosis. The clear understanding of its anatomical features and cranial foramina variants plays a critical role in finding the possible coping mechanism with its pathogenesis such as segmental instability or neurologic deficits.

Objective

This study aimed to investigate the incidence of occipitalization of Atlas and related variant foramina, as the baseline awareness of these conditions among the Nepalese population is yet to be documented.

Methodology

A retrospective study was performed for the total 86 dry skulls available in the department of Anatomy in Kathmandu University of Medical Sciences, Institute of Medical Science, and B.P. Koirala Institute of Health Sciences. The skulls were examined thoroughly to evidence the occurrence of cranio-vertebral variations.

Result

Out of 86 human adult skulls, 2 cases (2.32 %) were found with partial AOZ presenting posterior spina bifida close to the midline. Sphenoidal emissary foramen (SEF) was also observed in 17 skulls (19.76 %), an additional foramen lying anteromedial to the foramen ovale. Moreover, one of the skulls (1.16 %) was found with the presence of pterygospinous bar creating an additional foramen 'foramen of Civinini' in the lateral pterygoid plate of the sphenoid bone.

Conclusion

The incidence of AOZ and pterygospinous bar seems to be quite low as compared to the cases of SEF. However, the knowledge of such variations and the presence of additional foramina carry great significance for orthopedists and neurosurgeons to have prognostic implications and an accurate surgical approach.

KEY WORDS

Atlanto-occipitalization, cranio-foraminal variation, foramen of Civinini, pterygospinous bar, sphenoidal emissary foramen



INTRODUCTION

Occipitalisation of Atlas or the atlanto-occipital fusion is one of the skeletal variations where the adjacent part of the basiocciput gets fused with its succeeding first cervical vertebra (Atlas). In a normal skull, lateral masses of the atlas form ellipsoid synovial articulations with the occipital condyles. Since it is a biaxial joint, two movements are possible; one, around the transverse axis and the other, around the antero-posterior axis.¹ If there is the occurrence of atlanto-occipital assimilation, then there will be no existence of the aforementioned movements. Rather, it leads to atlanto-axial hypermobility and has been reported to be associated with atlanto-axial subluxation, basilar invagination, KlippelFeil Syndrome, and Chiari malformation.²⁻³

The basi-occiput and occipital condyles are developed from the caudal half of the fourth occipital sclerotome and the cranial half of the first cervical sclerotome. Whereas, the atlas or the first cervical vertebra is developed from the caudal half of the first cervical sclerotome and the cranial half of the second cervical sclerotome. During the fourth week of the embryonic period, the adjoining parts of sclerotomes proliferate so extensively that they get incorporated into each other, giving rise to an individual vertebra. However, the mesenchymal tissue lying intermediate to cranial and caudal parts of each sclerotome do not proliferate which eventually gets segmented by the development of intervertebral disc.⁴ The failure of segmentation between the cranial and caudal parts of the first cervical sclerotome leads to the AOZ since it gets assimilated with its preceding fourth occipital sclerotome. The fusion could be partial⁵ or complete,⁶ which might cause a major risk in neurovascular injury during the surgical intervention. In such cases, very careful examination is required in regard to the 4th part of the vertebral artery and the first cervical nerve. Besides, it also reduces the diameter of the foramen magnum⁵ that might cause compression in the spinomedullary region leading to neurological deficits.⁷ Knowledge of the normal ossification patterns is also necessary to find the variant cleft or foramina present in the skull.⁸

Thus, we aimed to investigate the incidence of occipitalization of Atlas and related variant foramina, as the baseline awareness of these conditions among the Nepalese population.

METHODOLOGY

The total 86 dry skulls were examined from the collection maintained in the department of Anatomy in Kathmandu University of Medical Sciences, Institute of Medical Science, and B.P. Koirala Institute of Health Sciences. This retrospective study was conducted during the period extending from Jan 2020 to March 2021. The skulls were examined thoroughly in order to identify the occurrence of crano-vertebral variations. The degree (partial or complete)

and the side of assimilation (unilateral or bilateral) were examined in the case of Atlanto-occipitalization. The anteroposterior (AP) and transverse (T) diameters of the foramen magnum (FM) were measured with the help of Vernier caliper. AP and T diameters of FM were measured as the maximum internal length along the midsagittal plane and the maximum internal width perpendicular to the midsagittal plane, respectively. These data were evaluated with the descriptive statistics using SPSS software version-25. Furthermore, the presence of cranial variant foramina was confirmed by inserting syringe-needle or probe through it.

RESULTS

Out of 86 adult human dry skulls, two cases (2.3 %) were found with the partial atlanto-occipitalization (AOZ). In both of the cases, AOZ was accompanied by the posterior spina bifida, close to the midline (Fig. 1-2). Lateral masses of the atlas were fused with the occipital condyles. One of those AOZ skulls presented a fusion of the right lateral mass of the posterior neural arch (Fig. 1B) with the posterolateral margin of the foramen magnum whereas another one had fusion on the left side (Fig. 2A). The anteroposterior and transverse diameters of foramen magnum were found with (34.5 ± 0.70) mm and (28.5 ± 0.70) mm respectively in the AOZ skulls, whereas the same measurements of the normal skulls were found with (35.5 ± 2.86) mm and (33.16 ± 2.7) mm respectively. Anteriorly, the neural arch of the AOZ skull was free from the occipital bone as in the normal skull (Fig. 1C). 'Sphenoidal emissary foramen' or 'foramen of Vesalius', an additional foramen lying anteromedial to the foramen ovale, was observed in 17 out of 86 skulls (19.7 %) (Fig. 3A). Moreover, one of the skulls (1.1 %) was found an interesting case of pterygospinous bar (Fig. 3B, shown with black arrow) present unilaterally on the left side. It was extended from the upper part of the lateral pterygoid plate to the spine of the sphenoid bone creating an additional foramen superiorly, known as pterygospinous foramen or 'foramen of Civinini' (Fig. 3B).



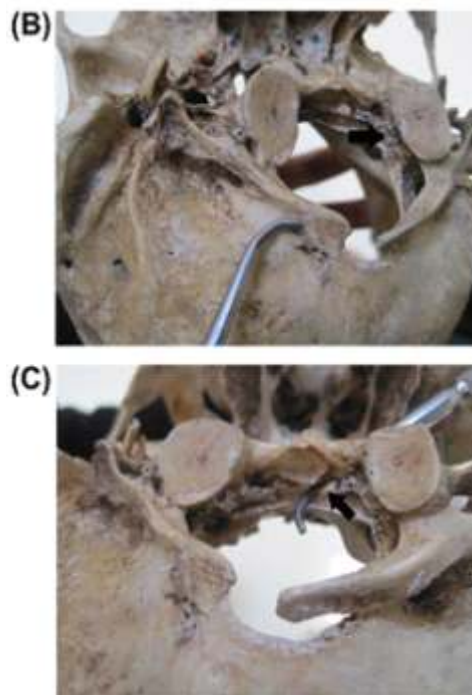


Figure 1 : Inferior view of Atlanto-occipitalized-skull. A) Spina bifida of Atlas is present approximately at the midline (shown with the black arrow). Posterior arch is free from basiocciput on the left side (shown with the probe). B) Lateral masses of Atlas are assimilated with the occipital condyles (shown with the black arrow). Posterior arch is assimilated to the basiocciput on the right side (shown with the probe). C) Anterior arch of Atlas is free from the basiocciput (shown with the black arrow).

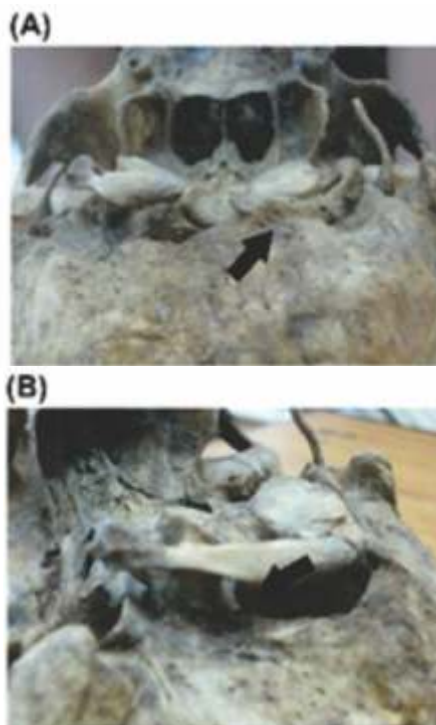


Figure 2: Inferior view of Atlanto-occipitalized-skull II. A) Lateral masses and posterior arch of Atlas are assimilated with basiocciput on the left side. B) Posterior arch on the right side is free (shown with the black arrow).

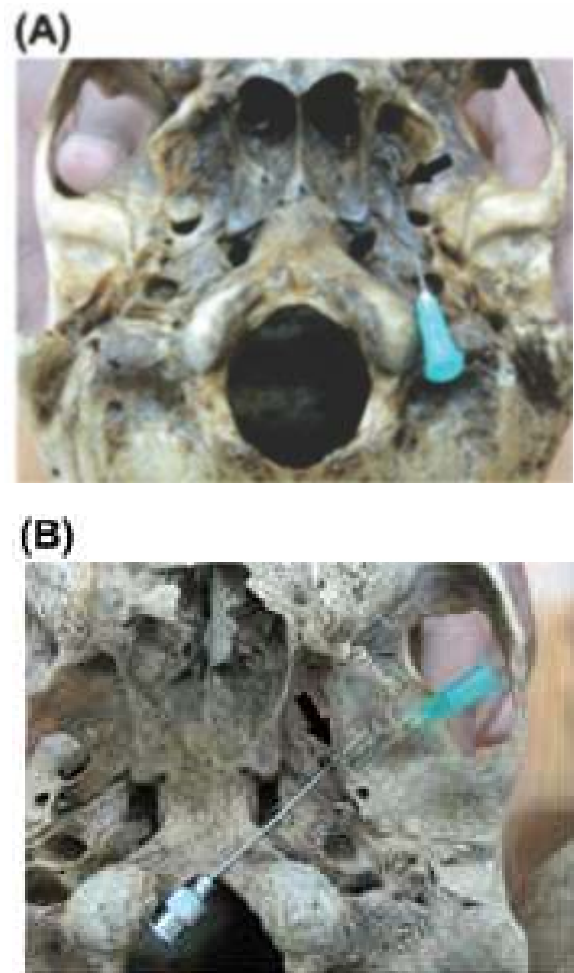


Figure 3: Inferior view of foraminal variant skulls. A) Sphenoidal emissary foramen or 'foramen of Vesalius' is present (shown with the black arrow). B) Pterygospinous bar (shown with the black arrow) creating an additional foramen above, called Pterygospinous foramen or 'foramen of Civinini' (shown with the probe inserting to it from the medial side).

DISCUSSION

Since the atlanto-occipitalization (AOZ) is associated with many other craniovertebral anomalies and the neurovascular pathology, the knowledge of its incidence is required to anticipate the clinical manifestation and their possible treatments. This study found the incidence of partial AOZ to be 2.3 % (2 out of 86 skulls) in Nepalese population (Fig.1-2). The review of it in the different populations ranges from 0.3 to 3.63 % (Table 1),^{5,9-12} the lowest in Thai,¹¹ and the highest in Malaysian populations.¹⁰ The AOZ is caused by the failure of segmentation between the cranial and caudal parts of first cervical sclerotome resulting the assimilation of the first cervical vertebra (the atlas) into the basicranium. It often leads to having the anomalous course of the vertebral artery that has to be seriously taken care of, during surgical procedures on the craniovertebral junction, hence, it requires well pre-



procedural planning with the help of computed tomographic angiography. In 78.4% cases of AOZ, the course of vertebral artery has been found passing through a bony canal between the fused part of the atlas and basicranium, whereas in 20.3% cases, passing below the atlas.¹³ The immobility of atlanto-occipital joint, on the other hand, causes hypermobility of lateral atlantoaxial joints and could overstress the transverse ligament which may lead to the atlantoaxial dislocation and the basilar invagination.¹⁴ The case of AOZ, associated with the reduction in the size of the foramen magnum (FM), may lead to the compression of the spinal cord and brain stem resulting in neurological symptoms. The standard dimensions for the normal FM vary between (28–38) mm for the sagittal diameter and (25–40) mm for the transverse diameter (Lang J, 1995) cited in (Skrzat J, 2010).¹⁵ The case of AOZ in our study showed only a slight decrease in anteroposterior diameter, whereas, a remarkable decrease in the transverse diameter of FM (Table 2). However, both reduction did not exceed the range of standard dimension, hence, might not have caused severe damage to the spinal cord.

Table 1: Review on Incidence of Atlanto-occipitalization (AOZ) among different population

Author, Year	Population	Incidence of AOZ No. of AOZ cases/Total skulls, %
(M. Sharma, 2008) ¹²	Indian	2/70, 2.8%
(Khamanarong et al., 2013) ¹¹	Thai	2/633, 0.3%
(Kassim NM & F, 2010) ¹⁰	Malaysian	2/55, 3.63%
(Natsis et al., 2017) ⁵	Greek	4/180, 2.2%
(A K., 2012) ⁹	Bulgarian	2/218, 0.9%
Shakya R., Present	Nepalese	2/86, 2.3%

Both cases of the AOZ were accompanied by the presence of spina bifida approximately at the midline (Fig.1-2). The case of only spina bifida usually remains asymptomatic unless there is any impingement on the surrounding structure including the spinal cord and vertebral artery. Such case of defect limited to the bone, seemingly normal as it is covered by the skin, is known as spina bifida occulta (SBO). Sometimes, the indications are visible on the newborn's skin showing abnormal tuft of hair or a small dimple.¹⁶ The SBO in Atlas is caused by the persistence of posterior midline synchondrosis between the two primary ossification centers of the posterior neural arch.¹⁷⁻¹⁹ The incomplete fusion of these ossification centers at the midline of the posterior neural arch may be normal for the age of 5-10 years.²⁰ The study on normal ossification pattern of Atlas showed that the complete fusion of ossification centers of the posterior neural arch occur at the average age of 5 years (age range 2-13 years).¹⁷ Some cases of atlas-SBO in the adults were found symptomatic of the cervicogenic headache (CEH) with the pain intensity; moderate (16 cases) and intense (1 case) out of 17 cases. In such cases, pain remission was observed after the blockade of the greater occipital nerve.²¹ However, there is a dispute regarding the atlas-SBO induced bilateral headache since many authors have described the unilaterality of the pain in the CEH.²²⁻²³

Apart from AOZ, some congenital anomalies of cranial base are associated with the presence of additional structures and foramina that holds the interest for clinicians where neurosurgical and vascular approach is a requisite. During embryogenesis, the cartilaginous precursors make its appearance to form the ala temporalis (future greater wing) from paraxial mesoderm, further growth of which, gets extended surrounding the nerves and vessels, creating the specific foramina for them. Sometimes, minor variations may take place resulting in the additional foramina, for e.g. 'Sphenoidal emissary foramen (SEF)' or 'Foramen of Vesalius'. It is believed that the SEF represents the site of fusion between, anteriorly ala orbitalis– membranous part, and medially ala temporalis–cartilaginous part.²⁴⁻²⁶ The present study found the case of unilateral SEF in 19.7% (17/86) skulls which seems to be a quite high incidence rate as compared to AOZ (2.3%, 2/86). But, our finding of SEF looks more or less concomitant to the data of previous studies in the Japanese (21.75%)²⁷ and the Turkish (28.1%, 81/317)²⁸ population. It has been found even higher in other populations ranging from 40–80% (Table 3),^{25,29-31} suggesting it as a common variation. However, its existence and consideration may help for the safer percutaneous approach to the middle cranial fossa through the foramen ovale, which could be mistaken with the SEF. Since it gives passage to a large emissary vein connecting the pterygoid plexus with cavernous sinus³², may cause profuse intracranial bleeding in case of its existential negligence.

Table 2: Morphometric measurements of Foramen Magnum in Nepalese dry skulls

Foramen Magnum	AOZ, n=2 (Mean ± SD) mm	Normal Skulls, n=84 (Mean ± SD) mm
Sagittal Diameter	34.5 ± 0.7	35.5 ± 2.86
Transverse Diameter	28.5 ± 0.7	33.16 ± 2.7

This study also found one interesting case of complete pterygospinousbar among 86 skulls (1.1%) present unilaterally on the left side. It is formed by the mineralization of pterygospinous (Civinini) ligament which is a thickening of interpterygoid aponeurosis, extending from the base of the spine of sphenoid to the posterior border of the lateral pterygoid plate. The published rate of pterygospinous complete mineralization is variable in different population with the range of 1.1 – 8.61% (Table 4).³³⁻³⁷ This bony bar is definitely a significant obstacle for neurosurgeons while approaching the retropharyngeal and parapharyngeal space.^{38,39} The complete mineralization of this ligament results in the formation of an additional foramen superiorly, 'pterygospinous foramen' or 'foramen of Civinini' (Fig.3B), very close to the foramen ovale, and giving passage to some branches of the mandibular nerve. In particular, lingual and inferior alveolar branches of the mandibular nerve may get compressed during the contraction of pterygoid muscles, developing a trigeminal neuralgia.⁴⁰



Table 3. Review on Incidence of Sphenoidal emissary foramen (SEF) among different population.

Author, Year	Population	Incidence of SEF No. of SEF cases/Total skulls, %
(Ginsberg <i>et al.</i> , 1994) ²⁹	American	99/123, 80%
(Kodama <i>et al.</i> , 1997) ²⁷	Japanese	87/400, 21.75%
(Ravalet <i>et al.</i> , 2015) ²⁵	Indian	90/150, 60%
(Loukas, 2018) ³¹	Greek	78/195, 40%
(Bayrak <i>et al.</i> , 2018) ²⁸	Turkey	89/317, 28.1%
(Leonel <i>et al.</i> , 2019) ³⁰	Brazilian	77/170, 45.2%
Shakya R., Present	Nepalese	17/86, 19.7%

Table 4: Comparison of Incidence of Pterygospinous Mineralization(PM) among different population

Author	Population	Incidence of PM No. of PM cases/ Total skulls, %
(Kapur E. <i>et al.</i> , 2000) ³⁴	Croats	4/305, 1.31%
(Nayak S.R. <i>et al.</i> , 2007) ³⁵	Indian	24/416, 5.76%
(Antonopoulou M. <i>et al.</i> , 2008) ³³	Greek	2/100, 2%
(Rosa R.R. <i>et al.</i> , 2010) ³⁷	Brazilian	8/93, 8.6%
Shakya R., Present	Nepalese	1/86, 1.1%

CONCLUSION

The incidence of atlanto-occipitalization and pterygospinous

bar seems to be quite low as compared to presence of an additional sphenoidal emissary foramen. The occurrence of craniovertebral synostosis and structural variations in cranial base, in terms of additional structures and foramina, bears a huge significance from clinical aspects. Hence, documentation of these variations among the Nepalese population, may contribute to prognostic implications, safer approach on surgery, or, broaden its therapeutic approach.

LIMITATION OF THE STUDY

Even though the dry skulls with the complete closure of fontanelles and completely ossified bones were considered in this study, there was no record of the exact age and gender.

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CONFLICTS OF INTEREST

The authors have no conflict of interest.

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