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Research article: Human Anatomy Case Report

Bilateral caroticoclinoid and absent mental foramen: rare variations of cranial base and lower jaw

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Summary

The human skull is a complex structure, which at birth is made up of 44 separate bony elements. Gross skull morphology and variations occurring in its important foramina (anomalous presence, absence, agenesis or multiplication) represent an interesting field of research, in order to achieve better comprehension of the embryological development of cranio-facial skeleton and the fate of its neuro-vascular content. Such anomalies may be asymptomatic in some cases while in others, they may carry important medical implications such as occurrence of atypical motor syndromes and influence the outcomes of modern neurosurgery. This case reports of an adult Asian female's dry skull that presented with two extremely rare foramen variations; the 'presence of bilateral caroticoclinoid foramen' and the 'bilateral absence of mental foramen'. To the best of author's knowledge, no similar case with co-existence of both variants in the same skull has been cited before in medical literature. This merits it's reporting as ostial variations of the cranial base and lower jaw hold diagnostic, surgical and developmental relevance for practicing clinicians, biological anthropologists and embryologists.

Key words

Anatomic variation, Caroticoclinoid foramen, internal carotid artery, clinoid process, mandible, mental foramen, mental nerve.

Introduction

The interior of the base of skull is divided into three fossae; anterior, middle and posterior cranial fossae. The anterior cranial fossa extends from the frontal bone anteriorly to the lesser wing of sphenoid posteriorly and lodges the frontal lobes of cerebrum. The lesser wings of sphenoid end medially to form eminences termed as anterior clinoid processes which are attached to the free margin of tentorium-cerebelli. The middle cranial fossa is butterfly shaped and is formed in the centre by the body of sphenoid. It presents the sella turcica whose boundary is completed laterally by two small eminences, the middle clinoid processes, which provide attachment for diaphragma sellae and form the medial boundary of the groove for the internal carotid artery.

The caroticoclinoid foramen (CCF) is an inconstant structure of the middle cranial fossa, formed by ossification of the fibrous interclinoid ligament or dural fold between the anterior and middle clinoid process. It allows passage of the interclinoid

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segment of internal carotid artery, as it turns upwards to supply the brain. Despite being an uncommon foramen, its knowledge is important owing to its location in the sphenoid bone, a neuro-surgically vital structure.

The mental foramen (MF) is an oval or circular opening on the anterior surface of the mandible; through which the inferior alveolar nerve and vessels, after passing through the inferior-alveolar canal, exit as the mental bundle and innervate the ipsilateral chin, lower lip and gingiva. The MF is the determinant of mental triangle and forms an important landmark of the human mandible.

In this case report we aim to describe multiple foramen variations in a dry human skull (presence of 'bilateral CCF' and 'bilateral absence of MF') and discuss their existence from a clinical and embryological perspective. Simultaneous occurrence of these anomalies is a unique morphological event with neuro-vascular and prognostic implications. Hence, they are worthy of note for clinicians as well as anatomists, should be kept in mind during skull/jaw diagnostic and surgical procedures and may help understanding cranio-facial development dynamics. Considering the fact that most anatomy textbooks do not provide a detailed description of the CCF and MF, the present report with its comprehensive literature review may prove especially relevant for orthognatic/neuro-surgeons and radiologists in day to day clinical practice.

Case report

During routine osteology demonstration, a dry skull (Asian/Indian female, 52 years) was identified in our anatomy museum as presenting with two rare foramen variations: presence of 'bilateral CCF' and 'bilateral absence of MF'. Records of the variations stated above are few and far between in medical literature and their parallel occurrence in the same skull is exceptional and merits reporting. Here, it is imperative to mention that bilateral absence of MF were cited earlier by the same authors (Hasan et al., 2010; available at http://www.ijav.org/pages/toc.html) in an independent case report while, at that time, the coexistence CCF remained undiscovered due to oversight by reporters. Radiographic scan of the average sized, partially edentulous skull showed normal bone density and absence of any invasive injury, geriatric bony resorption, fibrosis or osteo-pathology. This appears to be a case of developmental variance with ossification anomalies and congenital agenesis.

We do acknowledge that these foramen variations are embryologically and physiologically un-connected; the CCF results from anomalous ligamental ossification and has compressive effects on internal carotid vasculature while the absence of MF results from excavation failure of the inferior alveolar bundle and holds sensory-motor implications for lower jaw. Yet, how-so-ever unrelated these anomalies may be, awareness of their co-existence is a subject of academic research and clinical interest.

First variation: Bilateral caroticoclinoid foramen

The anterior and middle clinoid processes of both sides were linked by a thin bony spicule, thus forming complete CCF bilaterally (Fig. 1). The foramina were large and circular, with a smooth outline. They were located anterolateral to sella turcica, medial to the superior orbital fissure and behind the optic canal on both sides.

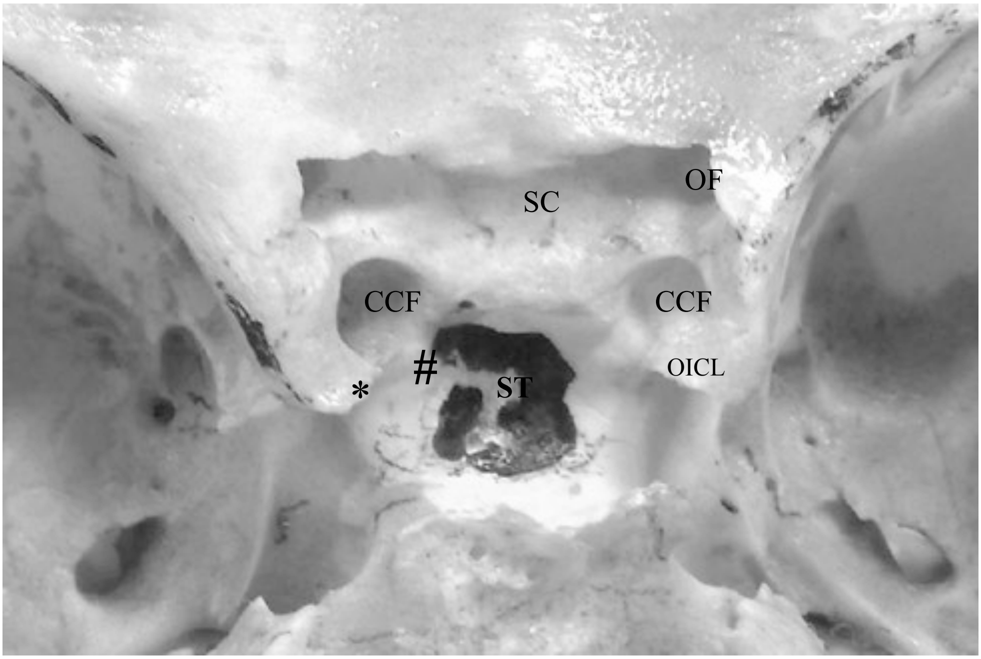

Figure 1 – Photograph of middle cranial fossa presenting with the anatomic variation of bilateral caroticoclinoid foramen (CCF). Also visible are optic foramen (OF), sulcus-chiasmaticus (SC), sella turcica (ST), anterior clinoid process (*), middle clinoid process (**#**) and ossified interclinoid ligament (OICL).

A manual caliper was used to measure maximum vertical and horizontal diameters and other topographically relevant distances for each foramen. To avoid errors, measurement was performed three times by the same examiner and repeated values were considered statistically. The maximum dimensions of the foramen were 5.23±0.52 mm horizontal and 5.15±0.44 mm vertical on right side and 4.8±0.44 mm horizontal and 4.6±0.59 mm vertical on left side. The length of the ossified carotico-clinoid ligament was 15±1.50 mm on right and 14.84±0.96 mm on left side and the average thickness was 1.7±0.88 mm on right and 1.81±0.75 mm on left side. For all these measurements, the difference between right and left foramen was statistically non significant ($p>0.5$).The anterior clinoid processes on the two sides were separated by a distance of 22.1±2.26 mm while the middle clinoid processes were 11.65±1.55 mm apart . The anterior and posterior clinoid processes were separated by a distance of 7.01 mm and 6.40 mm on the right and left sides respectively. The right middle clinoid process was prominent while the left one was rudimental. There was no tendency to fusion of the posterior clinoid process with either anterior or middle clinoid process.

Second variation: Bilateral absence of mental foramen

A careful visual inspection accompanied by radiographic scanning established that the mandible had normal gross morphology with absence of any signs of trau-

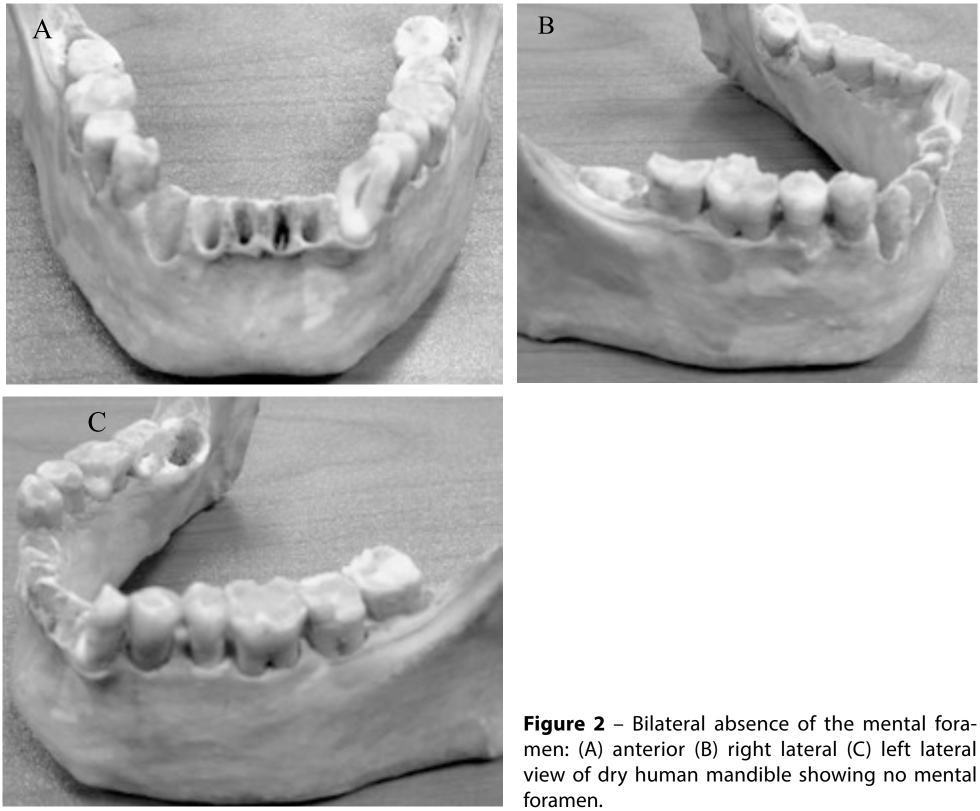

Figure 2 – Bilateral absence of the mental foramen: (A) anterior (B) right lateral (C) left lateral view of dry human mandible showing no mental foramen.

ma or pathology and normal bone density except for bilateral absence of the MF (Fig. 2A,B,C). Morphometry of related topographic landmarks was done by a manual caliper; the mandibular foramen were bilaterally symmetrical, measured an average of 33.55 mm on right and 32.82 mm on left side and was located 0.55 mm and 0.74 mm posterior to the centre of the ramus on right and left sides respectively. The inferior alveolar canals were postero-superiorly orientated with an average angle of 98.65° and was patent to a distance of 30.12 mm and 46.75 mm on right and left sides respectively as determined by a metal wire probe.

Discussion

Caroticoclinoid foramen

Although the fibrous ossification of ligaments is considered a normal physiological process that occurs with aging, this process is regarded as an 'exception' when pertaining to the formation of CCF which results from ossification of the interclinoid ligament or dural fold extending between the anterior and middle clinoid processes of sphenoid bone during embryological development. It was initially described by Henle (1855). Sellar bridges are laid down in cartilage at an early intrauterine stage and ossify in childhood. CCF are often observed in fetal and child skulls; in fact, a completely ossified CCF have been demonstrated in new-born infants (Hochstetter et al., 1940; Lang, 1995). Sellar bridges can cause several endocrinological and neurological problems during embryonic life. Being located very close to the hypophysis, they may cause pressure on the trochlear and abducens nerves that lie there, while passing laterally to the clinoid process. An interclinoid osseous bridge between the anterior and middle clinoid processes may cause pressure on the internal carotid artery that lies in the cavernous sinus and change the morphology of the terminal end of the bony arterial groove. Unilateral and incomplete CCF are a relatively more common occurrence (8 to 35%) than bilateral and complete foramina (0.2 to 4%) (Erturk et al., 2004; Freire et al., 2011). The incidence exhibits racial trends; occurrence is high among Turks, Portuguese, Nepalese and low among Brazilians, Koreans and Indians. The frequency of CCF has been observed as 9.9% in Japanese, 15.7% in Koreans, 17% in Alaskan Eskimos, 23.4% in Sardinians, 34.84% in Caucasian Americans , 6.27% in Portuguese, 14% in Germans and 35.67% in Turks (Erturk et al., 2004). It is also said that incidence of CCF is high (15–38%) in subjects with hormonal derangement, idiots, criminals, and epileptics (Lang, 1995). Perhaps its topographic position in the sellar region and its strategic relation to the hypothalamic-hypophyseal system may have some sort of correlation with such occurrence. Researchers quote equivocal findings on foramen dimorphism in relation to gender and right vs. left laterality; some report a female preponderance and no statistical differences in foramen occurrence between right and left sides (Lang, 1977; Erturk et al., 2004; Freire et al., 2011) while others report of foraminal predominance on the right side and a higher proportion in male skulls (Lee et al., 1997; Dodo and Ishida, 1987). The foramen observed in this case had an average diameter of 5mm; results similar to those reported by others (Dodo and Ishida, 1987; Erturk et al., 2004; Das et al., 2007; Haghanifar and Rokouei, 2009; Freire et al., 2011). CCF is a strategically important structure due to its relations with the cavernous sinus and its contents, internal carotid artery, sphenoid sinus and pituitary gland. Studies affirm that the position of CCF in an area close to the cavernous sinus may change dimensions of the intra cavernous area (Seoane et al., 1998). The degree of ossification in the foramen may vary from being ligamentous to partial or incomplete ossification to total ossification. Based on the extent of ossification, Keyers (1935) had classified the CCF into three types: complete, incomplete and contact type. There is a certain degree of correlation between the dimensions of CCF, the state of ossification of inter-clinoid ligament and the internal carotid artery; existence of a complete bony foramen may cause compression, tightening or stretching of the internal carotid artery because of attachment between dural rings of the artery and the interclinoid bridge of CCF. The CCF observed in this case were complete on both sides; such condition increases the possibility of vascular compression effects in the host subject. Studies have shown the presence of distinct morphological changes in the triangular clinoid space (Gupta et al., 2005) and internal carotid artery (Erturk et al., 2004; Das et al., 2007) in most cases of CCF. On one hand, a wide foramen may provide safety cover for the artery; on the other hand, it may also confuse radiologists while doing carotid arteriograms and pneumatization or marrow density assessments of the anterior clinoid process. During anterior clinoidectomy, CCF makes complete removal of the anterior clinoid process difficult and poses accessibility and hemorrhage problems during neurosurgical invasive procedures of the region like cavernous sinus exposure; especially in case of internal carotid or paraclinoid aneurisms and management of sphenoid and sellar-parasellar tumors. An encircling CCF may invade the caliber of internal carotid artery, giving rise to headaches and other compression symptoms owing to usual discrepancies between diameters of the artery and foramen; the former being larger in most cases (Das et al., 2007). However, to what extent this may possibly influence vascular biomechanics or produce functional disadvantage still remains a point for speculation. Insufficient flow dynamics may compromise circulation at the circle of Willis and its important vascular branches like cerebral and ophthalmic arteries. It may even impede important communications between the blood supply of fore and hindbrain, leading to inadequate cerebral perfusion and proneness to ischemia.

Unfortunately, most prominent anatomy textbooks fail to mention the CCF in sufficient detail, while this knowledge is valuable for radiologists and neurosurgeons. The presence of an interclinoid osseous bridge of CCF increases the risk of rupturing or tearing the internal carotid artery, leading to fatal cerebral infarction, especially when an aneurysm is present. It also makes it impossible to retract or mobilize the cavernous segment of the artery even after releasing the dural rings. Therefore, preoperative recognition of CCF by imaging has great clinical significance when approaching lesions in the clinoidal or sellar region. Comprehensive reports on CCF like ours can serve as an informative guide for neurosurgeons to recognize variations in the cranial base and reduce mortality and morbidity of surgical approaches in the region.

Mental foramen

It is an oval or circular opening on the anterior surface of the human mandible, through which the mental nerves and vessels exit to innervate the sub-mandibular or mental region. Mental foramen develops in the growing mandible, caused by the excavating action of the growing bundle of inferior alveolar nerve (branch of mandibular division of trigerminal nerve) and vessels. The neurovascular bundle traverses the mandible mesially, from lingual to buccal side. There is systematic bony resorption in the area, generating a tunneling effect resulting from programmed osteoclastic activity which creates a continuous channel for the growing nerve and vessels; which receives various names in sequence; the mandibular, inferior alveolar and mental canal. The morphology of the canal depends on the orientation, size, thickness, direction, angulation, growth rate etc. of the growing neurovascular bundle as well as the growth pattern of the developing mandible itself. The entry and exit points of the neurovascular bundle form the mandibular and mental foramen respectively. The direction of exit of MF on the buccal cortical plate of mandible is usually posterosuperior, with an average inclination of about 97° but other exit patterns like superior, labial, mesial and posterior also exist. A continuous bony canal is not always present between mental and mandibular foramina. It may lack definite walls, or end abruptly into multiple canaliculi. The MF undergoes changes in position and aspect during perinatal and post natal life, with a tendency for mostly haphazard displacement during early perinatal period and in a predominantly dorsal direction during late perinatal and post natal period. During the development of mental foramen, different phases of bone remodeling occur; apposition, inactivity and resorption. Separation of the neurovascular bundle into several fasciculi until the 12th week of gestation, earlier than the formation of MF, may result in accessory mental foramina while failure of tunneling activity by the neurovascular bud can lead to absent foramen which is an extremely rare phenomenon. There are discrepancies in shape, size, modal position and number of MF in different human races due to naturally occurring differences in facial skeleton, jaw structure, feeding habit induced bone remodeling of mandibles and relative differences between the growth of mandibular and alveolar parts of the bone. Available literature indicates that the average height and width of MF ranges between 3.3 and 3.6 mm (Haghanifar and Rokouei, 2009).The shape is predominantly oval and less commonly circular (Hasan, 2012). Variations in location of MF range from subcanine to submolar. The usual position of MF is below or between mandibular premolars; the most common location being below the second premolar (Hasan, 2012). Distinct racial trends have been observed in MF position; it is below the second premolar in Mongoloids, between the premolars in Caucasians and between the second premolar and first molar in Blacks (Santini and Land, 1990). Although MF location provides lesser taxonomic information than previously believed by paleoanthropologists, it is nevertheless considered a landmark for determining the maturity of human mandible and a highly suitable model to study the spatial relationship of nerves, vessels and dental primordia during bone remodeling. Quantitative diversity of MF (double/multiple/accessory or absent) holds important neuro-vascular implications pertaining to sensation around lower jaw and prognosis of invasive procedures around mental region. Supernumerary nerve fibers of accessory MF can reduce the effectiveness of inferior alveolar nerve block and surplus blood vessels can cause intraosseous hemorrhages during implant procedures (Sawyer at al., 1998). Accessory foramina are a far more common occurrence than MF absence; in fact, man is the only primate known to have agenesis of MF. Although reports of unilateral MF absence do exist in medical literature, its bilateral absence as cited here is an extremely rare incidence. An extensive search of published literature on prominent medical databases including Ovid Medline, Embase, Cochraine and Sciverse Scopus yielded just six cases of unilateral absence and a single case of bilateral absence between 1968 to 2012 (Table 1). Unlike accessory MF which demonstrate strong ethnic predisposition (Japanes > Melanese > American blacks > Greeks > French > American Whites > Russians / Asian Indians: range 1.5-12%; Sawyer, 1998), the incidence of MF absence does not exhibit racial trends. Apart from primary developmental agenesis, secondary factors accounting for absence range from post traumatic atrophy, fibrosis, osteoblastic hyperplasia, bony resorption or extreme positional displacements consequent of grossly misaligned ontogeny or geriatric receding of the alveolar margin of mandible that eventually consumes the mental foramen. Ambiguous radiographs account for a majority of cases of apparent absence and CT scans yield the best results, with minimum chances of errors in MF localization. The MF absence in this case report is most probably a result of bilateral congenital agenesis during mandibular development. This may cause neuro-sensory alterations in lower jaw in the regions supplied by mental nerve owing to lack of exit portal for the nerve; clinical implications could include ineffective mental block during anesthesia (Inke, 1968; De Freitas et al., 1979; Hasan et al., 2010). But evidence of whether or not sensorial alteration actually occurs remains inconclusive considering reports of living subjects with no sensory distur-

Table 1 – Variations in mental foramen number; Incidence of double, triple, multiple mental foramina (adapted from Hasan, 2012)

Author	Number of mandibles	Absent MF	%	Double MF	%	Triple MF	%	Quadru-ple MF	%	Year
Riesenfeld	3987			197	4.9					1956
Inke	1250	1 (unilateral absence)	.0001							1968
Azaz	105	1 (unilateral absence)	.01							1973
D Freitas	275	1 (unilateral absence)	.003							1976
D Feritas	1435	3 (unilateral absence)	0.2							1979
Gershenson	525			23	4.3	4	0.7	1	0.1	1986
Serman NJ	408			11	2.7					1989
Sawyer	705			42	5.9					1998
Stithipon	110			2	1.8					2005
Katakami	150			16	10.6					2008
Naitoh	157			11	7	2	1.2			2009
Hasan	NA*	1 (bilateral absence)	NA*							2010

*NA- Data not available in case report

bance despite MF absence. Da Silva Ramos Fernandes et al. (2011) have suggested that mental nerve and blood vessels in the region might be very thin but surely present, because the reported patients presented no innervation or vascularization disturbance in the mental and perioral region despite MF absence.

The MF is a morphologically and clinically important structure for anatomists, dentists and orthognatic surgeons. Erroneous localization can lead to in-correct prosthetic/surgical planning in the mental region. Violation of mental nerve can result in permanent paresthesia or anesthesia of the lower lip. Comprehensive knowledge of MF anatomy can significantly reduce the incidence of positional misjudgments in clinical dentistry and post surgical paresthetic, paralytic and hemorrhagic complications of the mental region.

The foramen variations described here are rare and may or not be frequently encountered during routine clinical procedures. Their subjects may remain a-symptomatic and un-diagnosed. However, knowledge of the same may create awareness and caution among practicing radiologists and surgeons, thereby decreasing chances of errors in diagnostic procedures, increasing the success and safety of invasive procedures, reducing pre-operative imaging misinterpretations, relieving irritating neurovascular symptoms, a-typical sensory-motor syndromes, functional impairment and post-operative morbidity. Supplementary information of such variants in routine anatomy text books would be a welcome change.

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