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CASE REPORT

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Two simultaneous anatomical variations of the cervical spine — a case report discussing the concept of tandem anomalies

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Background: *Arcuate foramen is an ossification of the posterior atlanto-occipital membrane, forming a bony opening through which the vertebral artery (VA) enters the vertebral canal. Block vertebra is a synostosis of at least 2 vertebral bodies that did not separate during the embryological development. It is worth distinguishing it from Klippel-Feil syndrome, as the latter oftentimes involves other abnormalities (namely skeletal) and is typically diagnosed in childhood. Both variants potentially lead to impairment of blood flow through the VA.*

Case report: *The following case report presents a finding of 2 anomalies of the cervical spine, found in a 38 y.o. female patient suffering from dizziness. A synostosis of the C4 and C5 vertebral bodies, arches, and zygapophysial (facet) joint was noted by the examining radiologist, with marked narrowing of the intervertebral foramen. Furthermore, a second anatomical variation in the form of the complete bilateral arcuate foramen was identified superior to the groove for the VA on the upper surface of the posterior arch of the atlas.*

Conclusions: *To the best knowledge of the authors, this case report is the first to present a co-existing block vertebra and bilateral complete arcuate foramen. The common presence of at least 2 anatomical variations that could have a synergistic clinical effect could possibly be termed 'tandem anomaly'. Notwithstanding, identification of a single anomaly explaining a patient's symptoms does not absolve the medical professionals from searching for any other potential variations that could also be present and could further influence the clinical picture.* (Folia Morphol 2025; 84, 1: 256–262)

Keywords: arcuate foramen, vertebral synostosis, vertebral artery, anatomy, radiology

INTRODUCTION

The cervical spine comprises the first 7 vertebrae (C1–C7). It is the most mobile part of the vertebral column and provides mechanical support to the skull. In addition to the aforementioned function, the foramina transversaria provide a route for the vertebral artery (VA) toward the posterior cranial fossa. The VA typically ascends from the subclavian artery and reaches the approximate level of the clivus, where the bilateral arteries merge into the basilar artery (BA). The clinical importance of the said blood vessels results from the vital role of neurological structures

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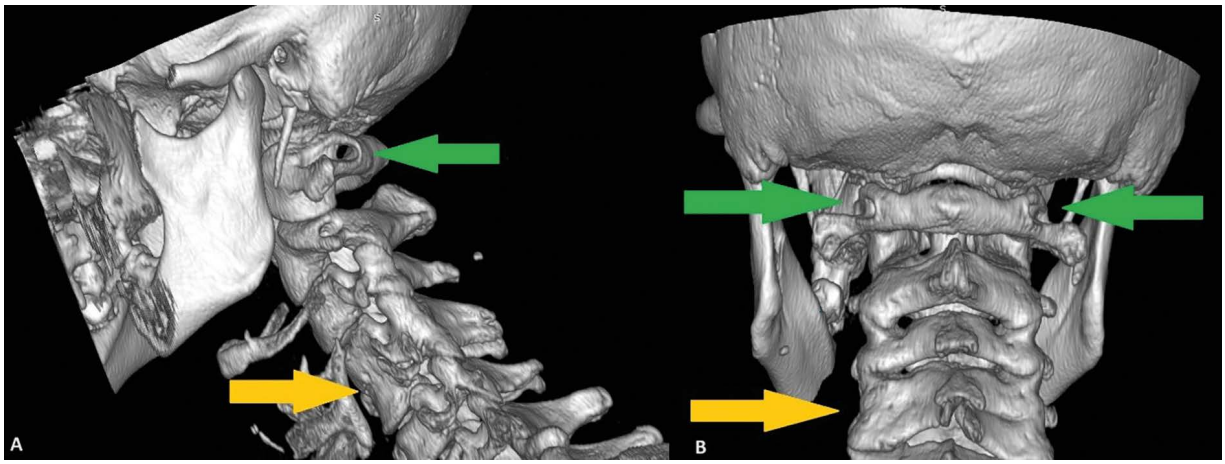

Figure 1. Lateral (**A**) and posterior view (**B**) of pseudo-3D reconstruction of the cervical part of vertebral column. Block vertebra (yellow arrow) and arcuate foramina (green arrow) are marked.

supplied by them, namely the brainstem and cerebellum [3]. Moreover, terminal branches of the basilar artery, i.e. posterior cerebral arteries, form the posterior ring of the circle of Willis, ensuring collateral circulation between the basilar and internal carotid arteries, to some extent [13, 16].

The arcuate foramen (otherwise known as the Kimmerle's anomaly or *foramen arcuale*, FA) is one of the most common among many anatomical variations involving the cervical spine. It appears when the lower border of the posterior atlanto-occipital membrane (PAOM), just above the groove for the VA, calcifies into the so-called *ponticulus posticus* — osseous bridge, which converts the groove into the foramen [5, 24, 29, 30].

The FA can radiographically be classified into one of 3 types based on classic radiograms: (1) the full type (with complete bony bridge transforming the groove into the foramen), (2) the incomplete (partially defective) type, and (3) the calcified type (characterised by linear or amorphous calcification in the atlanto-occipital membrane in the location of the FA) [22, 29]. Nowadays it is more common to use a simpler and less confusing way to categorise this anomaly. It is based on evaluation of the foramen, and not the ponticle itself, distinguishing the FA as complete or incomplete.

Data from a meta-analysis on the prevalence of the FA points to the relatively uncommon pooled prevalence estimate of its bilateral complete type in the general population [24]. Notwithstanding, co-existence of a yet another rare anomaly of the cervical spine, namely the block vertebra (BV) or vertebral synostosis, was the rationale behind the following case study.

CASE REPORT

The following study presents a unique tandem cervical spine finding, encountered during a computed tomography (CT) scan of a Polish 38 y.o. woman in the form of bilateral arcuate foramen accompanied by block vertebra at the C4/C5 level (Figs. 1–2).

The patient suffered from dizziness of unknown origin and was referred to a local Department of Medical Imaging by a consulting neurologist for a routine head CT scan in November 2022. The examining radiologist noticed a synostosis of the C4 and C5 vertebral bodies, arches, and zygapophysial (facet) joint. Because of the absence of intervertebral disc fusion of the adjacent vertebrae, the intervertebral foramen was noticeably narrowed, but only in the vertical diameter.

The transverse processes remained separated, as well as distal parts of the spinous processes of the involved vertebrae. Surprisingly, the fusion appeared to be asymmetrical — complete in the posterior and the left part of the vertebral body and arch, while those structures appeared to be slightly separated anteriorly and on the right side.

After a closer examination, a second anatomical variation in the form of complete bilateral FA was identified superior to the groove for the VA on the upper surface of the posterior arch of the atlas. There were no other deviations from the anatomical norm found in the CT.

DISCUSSION

During human ontogeny, somites that bilaterally develop around the neural tube give rise to myotomes (that will eventually lead to the formation of muscles)

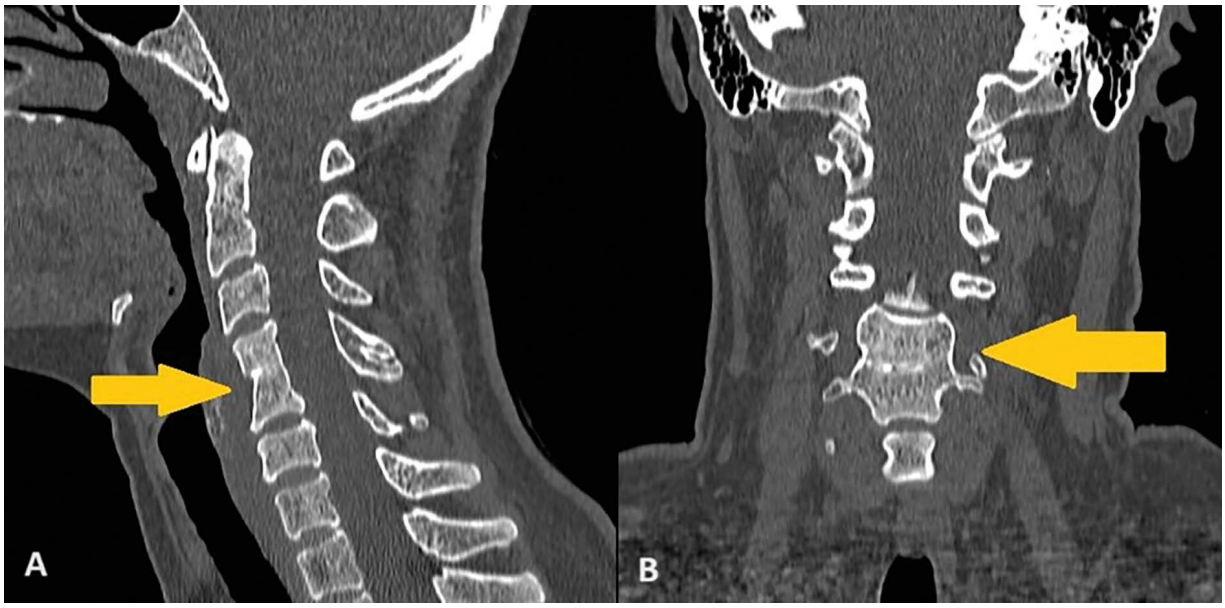

Figure 2. Sagittal (**A**) and coronal (**B**) view of the cervical part of vertebral column. Block vertebra (yellow arrow) is marked.

and sclerotome (that will eventually form the vertebrae) [23]. The primitive vertebrae seem to be separated one from the other by the presence of an intersegmental artery in a 3.5 mm foetus, with the segmentation best noted on paraxial sagittal sections [34]. The 3 stages of cervical vertebral development are regarded to occur as follows: (1) segmentation (week 3–6 of gestation); (2) chondrification (approximately week 6 of gestation) in the form of a single cartilaginous anlage; and (3) ossification (week 10–12 of gestation commencing from the posterior parts of the vertebrae, followed by the vertebral bodies 2–3 weeks later) [9].

The ligaments strengthening the atlanto-occipital and atlanto-axial joints, namely the PAOM, are formed by approximately the eighth week of gestation, and some of them (apical, alar, and transverse) provide a significant source of blood to the dens axis [9]. Lastly, the VA in the cervical spine (specifically the part within the foramina transversaria C1–C6) is a form of the post-costal longitudinal anastomosis between the cervical intersegmental arteries [10]. The VA originates from the seventh intersegmental artery (later forming the subclavian artery), whereas the portion of the VA that lies in its groove on the atlas is the spinal branch of the first cervical intersegmental artery [10].

The VA has been clinically divided into 4 parts, based on its relationship with the cervical spine. The first one (V1 — the pre-foraminal segment) constitutes the VA between its origin from the subclavian artery and the foramen transversarium (most commonly of the C6 vertebra). The second one (V2 — the foraminal segment) comprises the VA passing through the consecutive foramina. Having passed through the C2 vertebra, it forms the third segment (V3 — the atlantic, otherwise known as extradural or extraspinal). This part runs laterally to reach the transverse foramen of atlas. Next, the VA loops behind the atlanto-occipital joint, lying on the upper surface of the posterior arch of the C1 in its groove. The said arch is connected with the posterior margin of the foramen magnum by a broad ligament, the PAOM that separates the dura of the vertebral canal from the suboccipital muscles. Superior to the groove for the VA, the PAOM is pierced by the artery, thus entering the vertebral canal, accompanied by the first spinal nerve, leaving it using the same route. Having passed the membrane, the bilateral VAs travel up and reach the posterior cranial fossa via the foramen magnum, where they eventually merge into the BA. Hence, the fourth segment (V4 — intradural or intracranial) is located between the PAOM and the fusion of the bilateral VAs [1, 21, 35].

It is worth mentioning that the craniovertebral junction (comprising the cranial base, atlas, and axis, all connected with joints and strengthened by ligaments) is known to be highly variable [32]. Apart from the FA, incomplete posterior arch of the C1 or occipitalisation of the atlas into the skull are the most common examples. The latter is particularly

interesting in comparison to the FA because it can also impair blood flow through the VA at the same critical point, i.e. the transition from its extradural (V3) to the intradural (V4) segment [15, 31]. Few variants of possible route for the said artery have been described in literature in the case of atlanto-occipital assimilation. In about 60% of occipitalisations, the VA enters vertebral canal above the C1, through a foramen between the fused atlas and posterior margin of the foramen magnum, in the place corresponding to the location of the FA. In other cases, the artery may run underneath or behind the lateral mass of the atlas. In rare instances, it may be unilaterally absent [35].

A recent meta-analysis of 55,985 subjects found the pooled prevalence estimate of the complete FA to be 9.1% (95% CI: 8.2–10.1%), while the incomplete FA was present in 13.6% of cases (95% CI: 11.2––16.2%). Nonetheless, another important finding of the aforementioned study was that the lateral radiographs (modality used in older studies) were found to be the least sensitive in assessing the prevalence of FA compared to cadaveric studies and CT scans [24].

The same study stressed that it is not uncommon to find asymmetry in the structure of the FA. The complete type was identified bilaterally in about one-third of all cases (31.1%), the incomplete type was found contralaterally in 22.0%, and in the remaining 46.9% the complete FA was present only on one side with a simple groove for the VA on the other. Similarly, the incomplete type was found solely unilaterally in about half of the cases studied (52.3%), whereas the other half was contralaterally accompanied by the complete (16.5%) or the incomplete FA (31.2%) [24].

The BV is usually regarded as a congenital anomaly resulting from unsuccessful separation of adjacent vertebral bodies between the third and eighth week of gestation [7]. It is often attributed to the Klippel-Feil syndrome (KFS), which, apart from multilevel vertebral fusion, frequently involves other inborn abnormalities of the skeletal system and is hence in many cases diagnosed in childhood. In classical understanding of this disease, Klippel and Feil themselves proposed a triad of symptoms including limitation of motion, shortened neck, and low hairline [17]. However, in everyday clinical practice it has been observed that less than half of the patients present all those traits [7, 25]. Samartzis et al. [27] classified the KFS into 3 types, based on the distribution of congenitally fused vertebrae. Type I describes a single fused cervical segment; type II — multiple non-contiguous, fused segments; and type III — multiple contiguous, fused segments. It is crucial to remember that this classification applies only to congenital vertebral synostosis, because the fusion might occasionally be secondary to conditions like ankylosing spondylitis, juvenile rheumatoid arthritis, and trauma [7].

The estimated prevalence of KFS is 1 in 40,000 [33], although its diagnosis is based only on the aforementioned classification (especially since even solitary, one-level vertebral fusion meets the criteria of type I), the number may be as low as 1 in 17 [8]. Thus, it is why some authors insist on distinguishing between isolated congenital vertebral synostosis, i.e. BV, and KFS [18]. Regarding the presented patient, the authors are unaware of any other developmental anomalies normally attributed to KFS; hence, it was simply assigned as BV.

In the case of the described patient, both anatomical variants of the cervical spine could have affected the blood flow through the VA, collided with spinal nerves exiting vertebral column, or changed the range of motion of the spine. Possible consequences of both anomalies partially overlap and could be the reason behind the patient's dizziness.

It is believed that the FA may be responsible for a few conditions and symptoms, including cervicogenic headache, migraine, vertigo, and neck or upper limb pain. Most of those result from vertebrobasilar insufficiency [19]. Moreover, some data suggests that surgical resection of the FA may alleviate those problems [20]. It has been proposed that in unfavourable circumstances the FA may even be a cause of dissection of a vertebral artery and possibly lead to posterior circulation stroke [4]. Furthermore, it is worth remembering that the presence of FA may make it more difficult to plan and perform some surgical procedures designed to treat atlantoaxial instability, e.g. C1LMS — C-1 lateral mass screw. Potentially, misevaluation caused by mistaking the FA for the posterior arch of the atlas may decrease the safety of the procedure by putting the VA at risk of an iatrogenic injury [38].

The presence of the BV is also correlated with some pathological findings, including severe back pain, decreased neck mobility, and symptoms resulting from radiculopathy, namely sensory or motor deficits [6, 37]. Sometimes it may further affect the second segment of the VA and therefore produce symptoms related to its decreased blood flow, leading to, e.g., loss of consciousness triggered by neck

movement or position, in which the VA is constricted [14]. In a follow-up study of KFS patients who had undergone anterolisthesis, Alonso et al. [2] did not observe more frequent comorbidity of the adjacent vertebral segment to that of the congenitally fused segment. Nonetheless, longer follow-up periods and studies specifically dedicated to the BV are still warranted to elucidate this problem of possible adjacent degenerative spine disease for patients diagnosed with BV.

In cases of cerebrovascular pathologies, the term "tandem lesion" or "tandem occlusion" is commonly utilised in situations when circulation is impaired in 2 consecutive locations (most commonly the internal carotid artery and middle cerebral artery). The authors would like to adapt this term to describe co-existent anatomical variations involving the same structure, region, or functional unit as "tandem anomaly". The patient reported herein presented simultaneously those 2 abnormalities that together could be the cause of headaches, neck or back pain, vertigo, or even possibly stroke due to restriction of blood flow through the VA and therefore also the BA.

It is well known that certain conditions are more frequent in people with some anatomical predisposition. For example, there are authors [26, 36] who suggested that FA is present more often among patients who suffer from migraine headaches. Another meta-analysis has found that migraine headaches are significantly associated with an incomplete circle of Willis, especially when the anomaly is located in its posterior part [11]. Several studies analysing the structure of the circle of Willis found that some kind of incomplete variant is present in more than 80% cases, including not only its absence, but also hypoplasia, asymmetry, accessory vessels, or common origin of normally separate arteries [13, 16]. Assuming that more than 60% of the population has some kind of deficient component in the posterior part of the circle (dependent on the BA) [16], and bearing in mind that the FA is present in about 9%, it is probable that approximately 5% of the population might have those 2 anomalies, both potentially influencing the blood flow through the same parts of the encephalon. Even though such co-existent variants do not involve the same structure, they may both impair circulation in the same region; hence, they may be considered as a "tandem anomaly".

In the case of the patient presented herein, both findings applied to the anatomy of the cervical spine, and they may have negatively influenced the vertebrobasilar system, caused collision with spinal nerves, and potentially influenced the mobility of the spine. Thus, the probability of causing symptoms is much bigger in tandem anomalies in juxtaposition to isolated anomalies.

The main limitation of our study was the study type (case report), its retrospective character, and incomplete knowledge about the patient's full medical history. Future studies should pay more attention on combining results from both medical imaging and clinical perspectives, to discern the clinical significance of the spontaneously occurring variants. Moreover, studies on a larger number of participants might shed light on the true prevalence of various co-existing divergences from the anatomical norm in the cervical spine. The authors would like to acknowledge Sanchis-Gimeno et al. [28] in saying that clinical studies may potentially be biased by yielding a higher prevalence of variations (in their case of the AF), due to selection bias of symptomatic patients being examined, while clinically silent variants remain undetected.

The authors believe that more attention should be paid to the co-existence of multiple anomalies, and given their convergent clinical implications, such variants could possibly be termed as tandem anomalies.

CONCLUSIONS

To the best knowledge of the authors, this is the first case report to present a co-existing BV and bilateral complete FA. While it is common knowledge that every anatomical variant has its estimated prevalence and may be correlated with some pathologies, it is easy to forget that in rare cases 2 or more of those might be present in one patient. The authors would like to emphasise the fact that in some instances those anomalies could have a synergistic effect, for which the term 'tandem anomaly' has been proposed herein.

The authors would like to stress the importance of not neglecting further inspection of any other possible and especially related abnormalities, even after an anatomical variant that could be responsible for a patient's complaints has been identified. The former may also contribute to the patient's illness and wellbeing and could easily be overlooked if medical professionals concentrate too much on the first identified anomaly.

ARTICLE INFORMATION AND DECLARATIONS

Ethical statement

This type of study did not require approval from the local Ethics Committee. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Guidelines set by Henry et al. [12] applied.

Author contributions

Conceptualisation — JJ, BK. Project administration — IZ, BK. Methodology — JJ, IZ, BK, MPZ. Investigation — JJ, IZ, BK. Data curation — JJ, IZ, BK. Formal analysis — JJ, BK. Visualization — JJ. Writing (original draft) — JJ, IZ, MPZ, BK, JZ, ML, PP. Writing (review & editing) — IZ, BK, MPZ, JAW, JZ, ML, PP. Supervision — MPZ, JAW.

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Conflict of interest

The authors declare no conflicting interests.

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