Prevalence of the foramen arcuale of the atlas in a Saudi population

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ABSTRACT

الأهداف: وصف الخصائص الوبائية للثقبة القوسية لنقرة الأضلاع العمودية وانتشارها لدى المجتمع السعودي وعرض نظام تصنيف مبسط.

الطريقة: أجريت دراسة مقطعية في مستشفى جامعة الملك عبدالعزيز، جدة، المملكة العربية السعودية خلال الفترة من سبتمبر 2010م حتى فبراير 2011م. خلال مدة 6 أشهر تم إجراء 453 دراسة للعمود الفقري العنقي وتقييم درجات اختلاف التشوهات باستخدام نظام تصنيف طبي مناسب.

النتائج: أظهرت الدراسة أن 52.1% (236 مريض) ليس لديهم تجسر عظمي، و31.8% (144 مريض) لديهم تجسر عظمي خلفي غير مكتمل، و16.1% لديهم ثقبة قوسية للاضلاع العمودية بشكل تام. من خلال دراستنا ظهرت علاقة إحصائية فقط بين الجنس الذكري وحدوث التشوهات في الجانب الأيسر p=0.016 . المرضى المصابين بالتشوهات كن أكبر سناً من المرضى الذين لم تظهر لديهم أي تشوهات العمر=7.46 عام، p=0.034 . كما أظهرت هذه التشوهات ميلاً للأجزاء الجانبية الذي يعد بمثابة مصدر الاهتمام، k=0.592 القيمة الإحصائية=0.00 .

خاتمة: أن معدل انتشارهذا النوع من التشوهات في مجتمعنا مرتفع بالمقارنة مع الدول الأخرى الأمر الذي دعو إلى بذل المزيد من الفحوصات والدراسات.

Objectives: To describe the prevalence and morphologic characteristics of the foramen arcuale of the atlas vertebra in the Saudi population and propose a simplified classification system.

Methods: A cross-sectional hospital-based study was conducted at King Abdulaziz University Hospital, Jeddah, Kingdom of Saudi Arabia from September 2010 to February 2011. During the 6-month period, 453 CT studies of the cervical spine were evaluated for the presence of different degrees of this anatomic variant using a clinically relevant classification system.

Results: We found 52.1% (236 patients) to have no degree of osseous bridging, 31.8% (144 patients) had some degree of incomplete posterior osseous bridging, and 16.1% had the complete form of the foramen arcuale. The anomaly showed a male predilection that only reached statistic significance for those on the left side (p=0.016). Patients with a well-developed variant were older than those without the anomaly, but only by 7.46 years (p=0.034). These anomalies showed a propensity for bilaterality, which is a source for concern (kappa=0.592, approximate significance=0.00).

Conclusion: Compared to data from other countries, this anomaly has a higher prevalence in our population, indicating that further investigations are needed.

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The foramen arcuale is a poorly understood anomaly of the atlas vertebrae (**Figure 1**). Its origin, embryology, prevalence, clinical significance, and implications are all without definitive answers.[1] Records of its description have been found dating back to the 1800's.[2] Interest in this anomaly has since waxed and waned among spine surgeons, neurosurgeons, otorhinolaryngologists, neurologists, and chiropractors. It has been included

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in the differential diagnosis of numerous diseases and considered an indication as well as a contraindication for several surgical procedures.[1] Interest in this anomaly has now resurged with screw fixation of the atlas gaining favor in the spine community.[3]

A thorough review of the literature shows that the prevalence of this potentially morbid anomaly has not yet been described in our population. We describe the prevalence of foramen arcuale in a Saudi population, according to a clinically relevant classification system by Cederberg et al.[4]

Methods. We reviewed 453 consecutive CT scans that showed the craniovertebral junction from the foramen magnum to the bottom of the axis vertebra. The study was conducted at King Abdulaziz University Hospital, Riyadh, Kingdom of Saudi Arabia. The Biomedical Ethics and Research Committee approved this cross-sectional, hospital-based study as all imaging was indicated on clinical grounds and not specifically ordered for our study. We included studies in which CT scans were performed for imaging of the cervical spine, neck soft tissue, paranasal sinuses, and facial bones. We included patients who were above 20 years of age and had their studies carried out during a 6-month period from September 2010 to February 2011. Younger patients, or those with technically inadequate studies were excluded. These consecutive CT scan studies were from the university hospital's radiology database and not from specific services, a method we used to avoid selection bias. These included trauma cases, patients from the head and neck surgery, neurosurgery, and maxillofacial surgical services. Our radiographic database is digital, which allowed magnification of images on regions of interest and manipulation of contrast and brightness settings to best delineate anatomy. The literature was extensively reviewed, and the authors familiarized themselves with the various forms of this anomaly depicted in the literature, both cadaveric and radiologic. Various degrees of development were encountered and ranged from spicules to thick complete rings. We followed a simplified, clinically oriented classification system proposed by Cederberg et al,[4] which we believe is clinically relevant. It consists of 4 types, which are described in **Table 1** and illustrated in **Figure 2**. The importance of this classification is that it identifies what we believe are clinically and surgically relevant degrees of the anomaly (type 3, 4). These higher grades encircle most of the arterial circumference, which allows them to effectively tether the artery or act as sites of constriction. More rudimentary variants (types 1, 2) surround only a minority of the artery's circumference, and therefore should not affect its gliding motion on the posterior arch or serve as a source of confusion during dissection. All images were reviewed by one of the senior authors after initial screening by the 2 juniors involved in the study, with discrepancies resolved to insure consistent application of the classification system.

The data were entered and analyzed using the Statistical Package for Social Sciences (SPSS Inc., Chicago, IL, USA), version 17. The ethical review committee within the Surgical Department at King Abdulaziz University also approved the study.

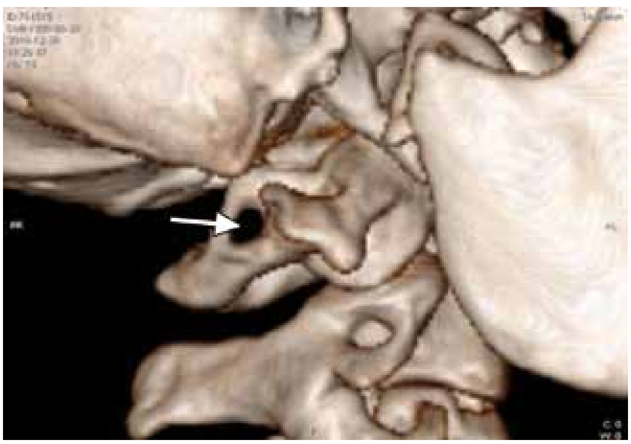

Figure 1 - Lateral view of a 3D-CT scan of a 21-year-old male demonstrating a complete type 4 form of foramen arcuale of the atlas vertebra.

Results. In our study sample of 453 patients, males accounted for 255 of the 453 patients (56.3%). The average age of our sample was 49.18 years (range 20-96), the mean age of males was 50.07 years, while that of females was 48.02, which is a statistically insignificant difference (t-test 1.32, degrees of freedom 451, and

Table 1 - Proposed classification of foramen arcuale of the atlas.[4]

Type	Description
1	**Normal:** No degree of this anomaly can be detected; including exaggerated grooving of the fossa arterialis with no distinct spicule.
2	**Trivial:** Partly developed foramen ranging from a minute but distinct spicule to a developing bridge that encases less than 50% of the circumference of the vertebral artery.
3	**Partial, well-developed:** Well-developed foramen encases at least the majority (greater than 50%) of the vertebral artery's circumference.
4	**Complete:** The vertebral artery is completely encased by bone.

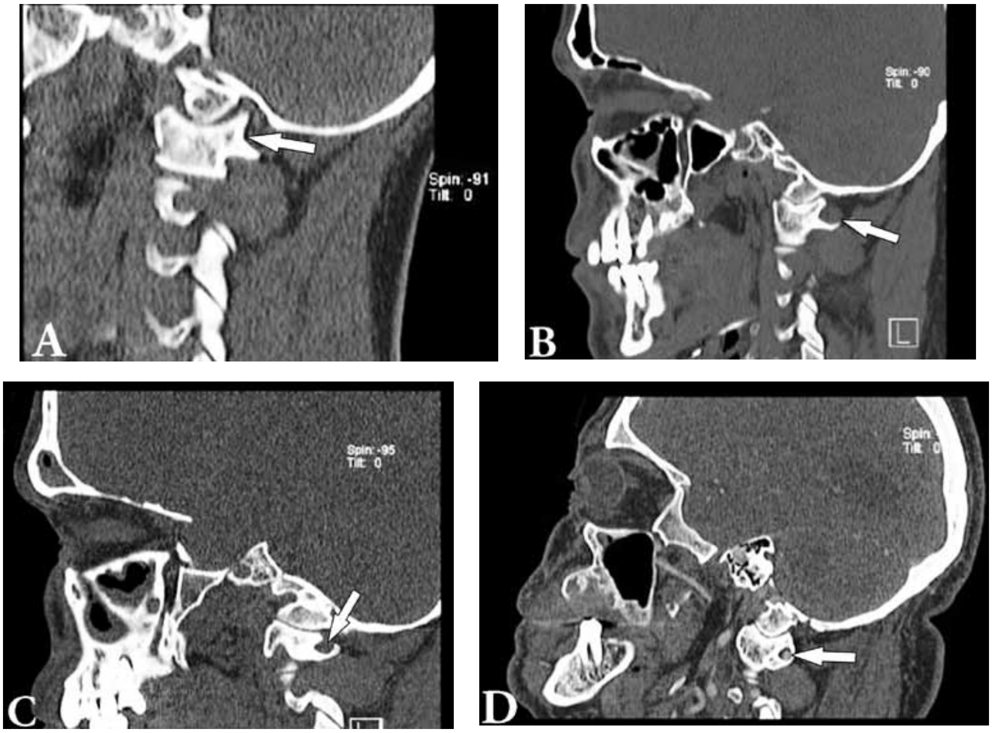

Figure 2 - Parasagittal CT scans demonstrating the classification of foramen arcuale: **A**) Type 1: Normal. **B**) Type 2: Trivial. **C**) Type 3: Partially, well-developed. **D**) Type 4: Complete.

p=0.187). The atlas vertebrae, when scanned for the osseous bridging, were normal (type 1) in 52.1% (236 patients), while 31.8% (144 patients) had some degree of incomplete posterior osseous bridging of C1 (types 2 and 3), and 16.1% (73 patients) had the complete form (type 4) of the foramen arcuale. Results are summarized in **Table 2**. When solely considering what we believe are clinically significant anomalies (types 3 and 4) that may serve as a tether to the vertebral artery, 27% (122 patients) of our sample had at least one side with a clinically significant anomaly (**Figure 3**). We also analyzed the sample for bilateral anomalies, since injuring a single vertebral artery intraoperatively may be tolerated, but bilateral injury can prove lethal. This was carried out using the Kappa measure of agreement to measure concordance between both sides (**Table 3**). Among the 122 patients with a type 3 or 4 variant, 50.8% (62 patients) had a clinically significant variant bilaterally (namely, both sides were either type 3 or 4) and therefore these patients would be at high risk for bilateral vertebral artery injury. We would also like to mention that left-sided anomalies are more likely to have a clinically significant contra-lateral side (Kappa value=0.592 and approximate significance=0.000) (**Table 3**). The anomaly showed a slight male predilection as the male to female ratio among those with some form types (II-IV) of osseous bridging was 1.15:1 (131 of 258 males [50.8%] compared with 86 of 195 females [44.1%]), the trend being consistent to varying degrees for all grades of the anomaly on both sides. Using the chi-square test, it had failed to reach statistic significance for the right side (*p*=0.1), but unexpectedly showed statistical significance for left-sided anomalies (*p*=0.016)

Table 2 - Observed laterality of different types of this anomaly (foramen arcuale) of the atlas.

Laterality	Complete type 4	Partial well-developed type 3	Trivial type 2
Bilateral	32	21	54
Right	28	18	22 with normal contralateral
Left	13	10	19 with normal contralateral
Total	73	49	95

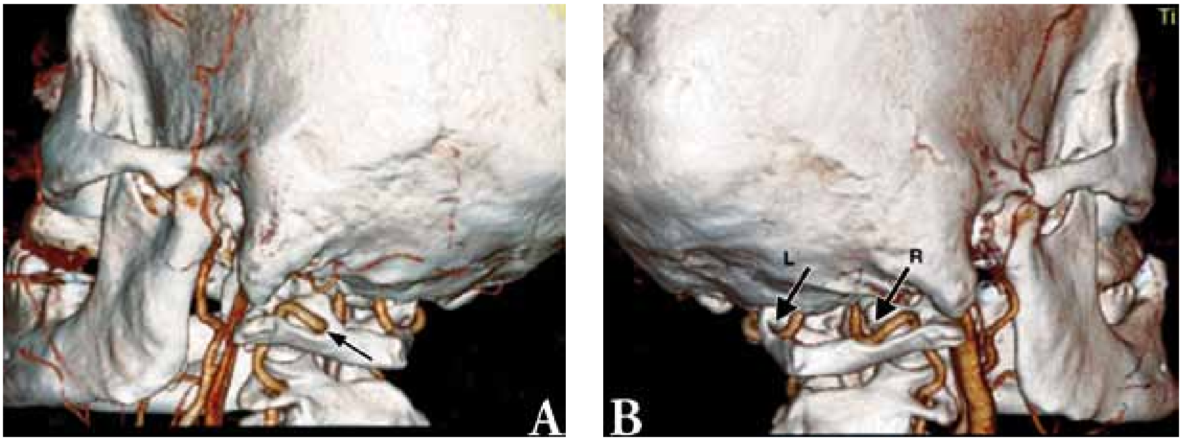

Figure 3 - The 3D-CT scans of a 38-year-old female with history of dizziness demonstrating **A**) complete type 4 foramen arcuale on the left side with the vertebral artery passing through (arrow), and **B**) the right side demonstrates type 2 variant consisting of only a bony spicule. L - left, R - right

(**Table 3**). We are without a satisfactory explanation for this observation.

The effect of age on the prevalence of the anomaly was investigated using a one-way ANOVA test (**Table 4**). The average age for anomalies were grade 1: 47.77, grade 2: 49.29, grade 3: 55.23, and grade 4: 51.00. A statistically significant difference in age was only found between grades one and 3 (p=0.034), but we find the difference not striking clinically, suggesting that the role of age may not be pronounced in its development.

Discussion. *Anatomical background.* As the vertebral artery courses from the transverse foramen of the atlas towards the foramen magnum, it lies on the posterior arch of the atlas in close relation to the posterior aspect of its lateral masses. In this part of its course, it is accommodated by a groove on the lateral masses, the fossa arterialis (sulcus arteriae). In some instances, an osseous bridge extends from the posterior aspect of the superior articular process towards the posterior arch of atlas. When fully developed, it converts the artery's groove into a foramen, the foramen arcuale.[1]

This anomaly is not new to the anatomists, having been attributed to Kimmerle in 1930.[5] However, descriptions of this anomaly have been found to date back to the 1800's by Allen.[2] Many different synonyms exist and can be a source of confusion to the reviewer, these include the posterior ponticle, ponticulus posticus, arcuate foramen, foramen sagittale, foramen atlantoideum posterior, Kimmerle's variant, foramen arcuale, foramen retroarticular superior, canalis vertebralis, retroarticular vertebral artery ring, retroarticular canal, and retrocondylar vertebral artery ring.[3]

The origin of this anomaly is controversial. Some claim it to be congenital, citing cadaveric and radiologic studies that have shown its presence in fetuses and children, with some still in the cartilaginous stage yet to ossify.[6] They suggest its origin as either a remnant of the proatlas or the result of primitive ligament

Table 3 - Measure of agreement (Kappa test) between the right-sided and left-sided anomalies (K value=0.592)

Anomaly	Left-sided anomaly		Total
	Type 3 & 4	Type 1 & 2	
Right-sided anomaly			
Type 3 & 4	62	41	103
Type 1 & 2	19	331	350
Total	**81**	**372**	**453**

Table 4 - Mean age of various degrees of the anomaly (p-value between groups=0.034)

Anomaly	Number	Mean	95% CI
Type 1	267	47.77	45.90-49.64
Type 2	83	49.29	45.41-53.16
Type 3	43	55.23	50.56-59.90
Type 4	60	51.00	46.45-55.55
Total	**453**	**49.18**	**47.68-50.69**

ossification.[7] An alternative theory suggests that it may be due to degenerative calcification of the posterior atlantooccipital membrane.[7] This "acquired" theory gains support from the noted increase in prevalence among laborers exposed to regular axial loading (as in carrying objects on the head), indicating that a degenerative element has at least some role in its development.

Prevalence. The prevalence of this anomaly in the literature varies widely, with reports ranging from 1.14-37.8%.[1] The methodology of these studies is inconsistent, with most being cadaveric, and the remainder consisting of an inhomogeneous assortment of lateral cephalograms, cervical spine x-rays, and cervical spine CT scans. It has been shown that simple radiographs can miss these variants, and are limited in their capability to provide information on their bilaterality.[8] A recent study found that among the same population, CT's had a statistically significant higher yield for this variant when compared to a technically adequate lateral radiograph.[8]

We noted a paucity of studies on the prevalence of this anomaly in neighboring populations, with only 2 studies conducted on the Turkish population. The first having showed a prevalence of 7.2% for the complete form, and 6.25% for the incomplete forms after reviewing over 400 lateral cervical radiographs, although the same study also describes a prevalence of 11% for the complete form, and 3% for the incomplete variant in a cadaveric survey of 60 atlases.[9] The other Turkish study mirrored the results of an Iranian survey that revealed a much lower prevalence of this anomaly hovering around 5%.[10,11] Our results reveal a high prevalence of this anomaly in our population. The figures we present are higher than those reported in the literature describing the prevalence of this anomaly in neighboring countries.[9-11] We do not have a definitive explanation for this observation, but we are one of the very few studies that utilized CT as our study material, and it may be that the higher yield of this modality is the cause.[8] Our results show the limited effect of age on the prevalence of the anomaly, which is echoed elsewhere.[4]

Diagnostic imaging. The diagnostic method of choice to detect this anomaly is a CT study, with CT vertebral angiograms and 3D reconstructions as useful adjuncts.[8] They allow the physician to not only assess the atlas, but also assess whether stenosis of the third (V3) segment has occurred in the tunnel. Other modalities that may be investigated as adjuncts in decision-making include Doppler velocimetry and dynamic angiographic studies to assess vertebral blood flow at varying degrees of neck rotation. These may prove especially useful to the surgeon and anesthesiologist in assessing the effect of the stressful intraoperative positions needed for some surgeries, and to the physician encountering a patient with paroxysmal symptoms of vertebrobasilar insufficiency and apparently normal vertebral arteries.

Clinical significance. Knowledge of the structures that pass through this foramen is essential to understanding the attributed significance. Accompanying the vertebral artery is the venous plexus, periarterial sympathetic plexus, suboccipital, and first cervical nerves. This anomaly affects these structures either by compressing them or acting as a tether, preventing their normal gliding motion in the posterior arches of the atlas as the neck rotates.

The aspect relevant to neurological and spine surgeons operating on the atlas lies in its tethering effect on the V3 segment of the vertebral artery. This has potentially catastrophic consequences in a variety of procedures: a breach of the superior cortex during a simple C1 laminectomy; V3 segment mobilization in far lateral skull base exposure; or screw insertion into the posterior arch will encroach on this canal resulting in arterial compression, injury with bleeding or dissection and thrombosis. Taking into account that a partially exposed ponticulus in the operative field resembles a broad posterior arch of atlas as illustrated in **Figure 4**, may result in a dire screw trajectory that traverses the vertebral artery. The risk is compounded by our observed bilaterality of this anomaly. This is very important to address, since screw fixation of the atlas is gaining favor in the spine surgical community.[12,13] No reports have surfaced of vertebral artery injury arising as a consequence, but there have been reports of instrumentation being deferred because

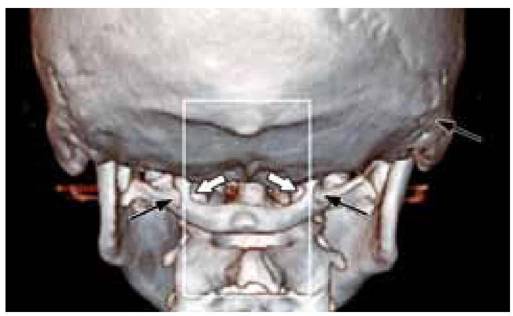

Figure 4 - The 3D-CT scan posterior view of a 32-year-old male demonstrating bilateral type 4 foramen arcuale. Notice the widening of the posterior arch laterally with a great possibility of having inadequate atlas exposure (white box) and injury of the vertebral artery if instrumentation is attempted.

of intraoperative identification of this variant and lack of experience in addressing this anatomic obstacle.[14] Since the consensus now is modification of the screw trajectory to avoid injuring the vertebral artery, it is not to be regarded as a contraindication for screw fixation of the atlas.[15] We believe that carefully extending the dissection laterally to fully expose the arcuate foramen and untethering the artery may provide a solution if no other option is feasible.

The anomaly's operative importance, however, is not restricted to surgeries on the craniovertebral junction. A recent report[16] attributed an intraoperative vertebrobasilar stroke, proposing that neck hyperextension intraoperatively may have kinked the artery tethered by the osseous ring. That incident altered the intraoperative positioning guidelines at that institution for patients identified as having this anomaly. This makes it relevant to both the surgeon and anesthesiologist.

By virtue of the number of structures passing through this foramen, many other symptoms and syndromes have been linked to this anomaly in the literature. These include headache from compression of the first cervical nerve, vertigo from vertebral insufficiency, Wallenberg syndrome, and the aptly named Bow Hunter's stroke from its occurrence at the extremes of lateral neck rotation.[17-20] Other poorly understood entities attributed to the arcuate foramen include: the syndrome of Barre-Lieou, tongue fasciculations, vertebrobasilar insufficiency, and dysphasia.[21,22] Hypothesized to be a risk factor, one case series identified the anomaly in 9 out of 12 children with traumatic subarachnoid bleeds of the posterior fossa.[23] Several reports of symptom improvement after decompressing this canal indicate its possible role in causing disease and that it may also be an unidentified culprit in the etiology of numerous conditions thought to be idiopathic.[18,21-24] This is in stark contrast to a study questioning the relevance of this anomaly, displaying minimal effect on vertebral artery dynamics with neck rotation.[25]

The displayed high prevalence of this anomaly in our study and its as-of-yet uncertain but potentially morbid significance make it a relevant target for local research to further describe its clinical importance and true prevalence. Our study is limited by the lack of clinical correlation, as we have not questioned and examined these patients for relevant signs and symptoms. Nor have we assessed the vertebral artery by dedicated studies. Our aim was to merely establish the prevalence of the arcuate foramen in our population and reveal whether it warrants further investigation.

In conclusion, posterior osseous bridging of the atlas is fairly common in our population. It seems that a definitive statement on the clinical importance is yet to be offered by the literature. Most mentions of this anomaly in the literature are in the form of case reports and series, with high quality evidence severely lacking. We report a study assessing the prevalence of this anomaly in our population. We hope to have raised awareness of this potentially significant anomaly in our community in the hopes that further studies will provide us with much needed answers about its significance both as a cause for symptoms and operative complications.

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