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Clinical Study

Hypertrophy of the anterior arch of the atlas associated with congenital nonunion of the posterior arch: a retrospective case-control study

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Abstract

BACKGROUND CONTEXT: Nonunion of the posterior arch of the atlas is an uncommon but normal developmental variant. It is usually asymptomatic in the patient but may be associated with greater incidence of fracture because of increased stress on the anterior arch.

PURPOSE: We sought to determine whether anterior arch hypertrophy is present in cases of congenital nonunion of the posterior arch of the atlas.

STUDY DESIGN/SETTING: A retrospective analysis of 1 year (February 2005–January 2006) of computed tomography cervical spine studies requested by the University of California San Diego Medical Center Trauma Department was undertaken.

PATIENT SAMPLE: All patients matching the search criteria (see *Study design*) were included.

OUTCOME MEASURES: Area density product, defined as the midline cross-sectional area of the anterior arch on sagittal reformat multiplied by the average areal radiodensity in Hounsfield units (HU) as measured by two raters, was calculated for cases and controls.

METHODS: Cases of posterior arch nonunion were identified and matched to controls. The significance of differences in area density product between cases and controls were established by the Student *t* test. Interrater correlation was calculated.

RESULTS: Posterior arch nonunion was identified in 26 individuals (3.1% of 839 studies reviewed). Compared with age- and sex-matched controls, a 21% increase in area density product of the midline anterior arch was observed in posterior arch nonunion cases (773 HU-cm^2 in cases vs. 637 HU-cm^2 in controls; $p<.001$). This increase was attributable to a 21% increase in cross-sectional area (1.05 cm^2 in cases vs. 0.87 cm^2 in controls; $p<.002$). In contrast, there was no significant difference with regard to increased average radiodensity.

CONCLUSIONS: It has long been subjectively recognized but not objectively quantified, until the present study, that the anterior arch of the atlas is hypertrophied in cases of posterior arch nonunion. Anterior arch hypertrophy may represent an adaptive response to chronically elevated mechanical stress and loss of hoop strength in cases of posterior nonunion.

Keywords: Cervical atlas; Congenital; Spine; Spinal fractures; Radiology; Retrospective Study

Introduction

The atlas (C1) is the topmost vertebra. Its ring-like structure is composed of an anterior arch, two lateral masses, and a posterior arch. It supports the skull superiorly, and its odontoid articulation with the axis (C2) affords the cervical spine its liberal range of rotational motion.

During development, the atlas classically ossifies in three centers. The two lateral centers appear in the seventh week and form the lateral masses, which also extend posteriorly to form the incipient posterior arch. At birth, a narrow segment of cartilage separates the two posterior hemiarches; fusion occurs at age 3 to 5 years. The primordial anterior arch is fibrocartilaginous at birth, the developmental remnant of the hypochordal bow. The third center of

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ossification appears anteriorly at the end of the first year of life, eventually forming the mature osseous anterior arch, which fuses with the lateral masses at age 5 to 9 years.

Nonunion of the posterior arch of the atlas is an uncommon but normal developmental variant. Failure of midline fusion of the two posterior hemiarches accounts for the majority of such defects, with prevalence estimated at 1% to 4.3% of the population based on cadaveric dissections [1] and retrospective survey of neck and cervical spine computed tomography (CT) scans [2]. Non-midline nonunion of the posterior arch is less common, and anterior arch nonunion is exceedingly rare [3]. In the emergency department, it is important to distinguish variant anatomy from acute trauma [4–9], that is, Jefferson fractures [10–12] and odontoid fractures [13,14], based on criteria such as cortication, soft-tissue swelling, lateral offset [12], and/or cervical spine instability during clinical examination [15].

Most atlantal defects are present in asymptomatic patients and represent normal variants [3]. Less commonly, posterior arch abnormalities can have severe neurologic deficits when associated with conditions such as Arnold-Chiari malformation, gonadal dysgenesis, Klippel-Feil, Turner, and Down syndromes [16]. Atlantal defects are associated with greater degree of atlantoaxial instability and susceptibility to injury after minor head or neck trauma [17–19]. It has also been suggested that atlantal defects are associated with compensatory changes such as hypertrophy of the anterior arch of C1, hypertrophy of spinous process of C2, and hypertrophic downward projection of the posterior border of the foramen magnum [17].

We hypothesized that in cases of congenital nonunion of the posterior arch, additional mechanical stress would be exerted on the anterior arch; hence, the anterior arch would adaptively remodel in these individuals and achieve greater cross-sectional area, greater radiodensity, or both, compared with age- and sex-matched control patients.

Material and methods

Institutional review board approval was obtained for retrospective review of 1 year of CT cervical spine studies ordered by the University of California San Diego Medical Center Trauma Department from February 2005 to January 2006 inclusive, using Agfa picture archive communications systems (ie, PACS) search criteria. Initial exclusion criteria were: Jefferson fracture, digitized outside images, and lack of age identifier.

Age, sex, and the presence of an open or closed posterior arch were recorded for each study. If a posterior arch cleft was present, area density product (ADP) was calculated on standard 1-mm thickness midline sagittal reformat image. ADP was defined as the midline cross-sectional area of the anterior arch on sagittal reformat image, measured by free-form mark up in Agfa PACS and multiplied by the average areal radiodensity in Hounsfield units (HU) measured on this same image that had been magnified 15 times on a window width/level of 2000/500. These same measurements and calculations were then performed on normal age- and sex-matched control patients who had no cervical spine abnormalities. If anterior arch ADP could not technically be obtained, the case-control pair was excluded. Means of anterior arch midline cross-sectional area, average areal radiodensity, and ADP values for cases and control patients were compared. Statistical significance was established by a paired Student t test with a two-tailed distribution. All measurements were performed by two radiologists, who were both board-certified and fellowship-trained in musculoskeletal radiology. Interrater correlation was calculated.

Results

By search criteria, 847 CT scans were found. Eight scans met initial exclusion criteria. In total, 839 scans comprising 623 male and 216 female patients were reviewed. The ages ranged from 11 to 99 years; average age was 41 years.

Nonunion of the posterior arch (Fig. 1) was noted in 26 individuals (3.1%) with an age range of 17 to 87 years, and an average age of 43 years. Twenty of these patients presented with a simple cleft; the remaining six individuals comprised 1 split atlas, 1 near-complete occipitoatlas fusion, and 4 unilateral or bilateral occipitoatlas fusions. The first two individuals were excluded from analysis because a midline anterior arch cross-sectional area could not be measured. One case of anterior arch stress fracture was observed; posterior nonunion was noted in this same individual (Fig. 2).

Anterior arch ADP means, cross-sectional area, and average areal radiodensity, averaged across both raters, were

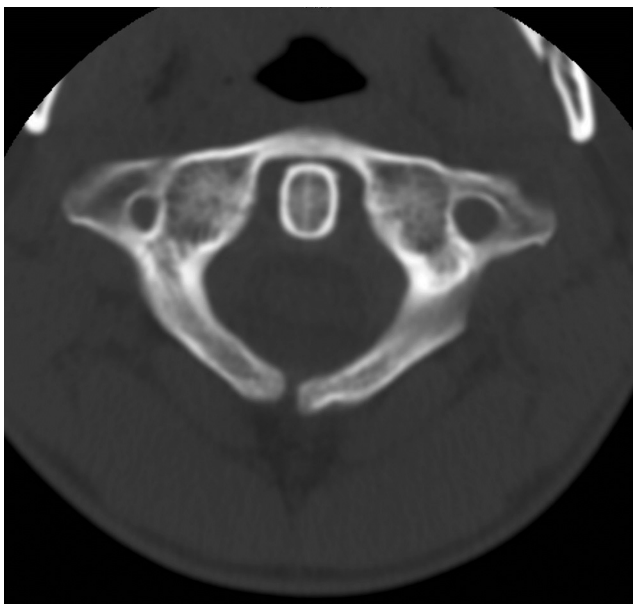

Fig. 1. Axial view of CT cervical spine at the level of C1: nonunion of the posterior arch. CT, computed tomography.

Fig. 2. Axial view of CT cervical spine at the level of C1: a stress fracture of the anterior arch of the atlas with associated posterior nonunion. CT, computed tomography.

obtained in both posterior arch nonunion cases and in control patients (Fig. 3), with percentage change in cases versus controls, statistical significance, and interrater correlation noted (Table). There was a 21% increase in both ADP ($p<.001$) and cross-sectional area ($p<.002$) in cases versus control patients. There was no statistically significant difference in average areal radiodensity.

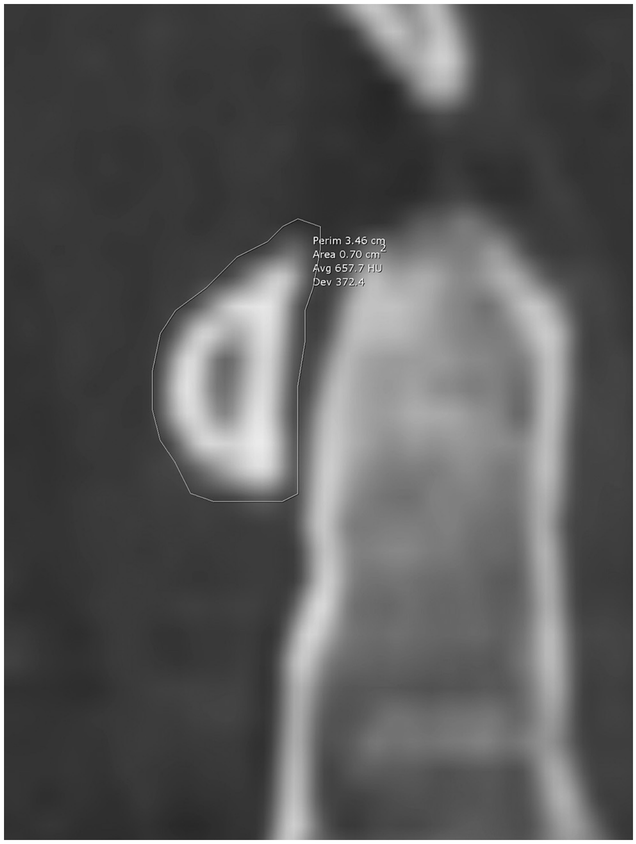

Fig. 3. Sagittal view of CT cervical spine at the level of C1: measurement of midline cross-sectional area and average areal radiodensity is shown. CT, computed tomography.

Discussion

Posterior arch nonunion variant was seen in 3.1% of CT cervical spine studies (839 total scans) requested by the University of California San Diego Medical Center trauma department. The incidence of this variant is consistent with previously performed cadaveric dissections and retrospective CT cervical spine surveys, which ranged from 1% to 4.3% [1–3,20]. Although uncommon, the incidence is high enough to warrant clinical recognition. Compared with age- and sex-matched control patients, a 21% increase in the ADP of the midline anterior arch was observed in cases of posterior arch nonunion (773 HU-cm^2 in cases vs. 637 HU-cm^2 in control patients; $p<.001$). Anterior arch hypertrophy in posterior nonunion suggests an example of Wolff's law: adaptive bone remodeling in response to chronically elevated mechanical stress.

Interestingly, the increase in ADP was attributable to a 21% increase in cross-sectional area of the anterior arch (1.05 cm^2 in cases vs. 0.87 cm^2 in controls; $p<.002$). In contrast, there was no significant difference with regard to increased average radiodensity, a finding also is in concert with previous experimental work, which demonstrated that although mechanical stress resulted in increased cortical thickness and therefore cross-sectional area, it did not change the mechanical properties of bone [21]. The lack of change in average radiodensity suggests that the fundamental trabecular density and composition of the anterior arch are not significantly changed. Bone quantity is increased, and bone quality is conserved.

However, the increase in quantity of bone does not completely negate the fundamental loss of hoop strength due to posterior arch nonunion. Hence there may be greater risk for anterior arch fracture or stress fracture if the increased motion and concomitant stress facilitated by posterior arch nonunion is excessive [20,22,23]. Tellingly, the single case of anterior arch stress fracture in our study (0.1% of 839 scans) was noted to be present in an individual with a posterior arch nonunion. However, firm conclusions cannot be drawn based on a single case.

This study had several limitations. The study population was limited to trauma patients with CT cervical spine imaging available. The number of cases was not large enough to do stratified analysis on the basis of age or sex, although these factors were controlled for to reduce confounding. There was some rater variance in determination of the midline sagittal image, and cross-sectional area and average areal radiodensity were based on freeform markup. Although interrater correlation was very high, intrarater reliability

Table
ADP in cases versus controls

Measure	Cases (n=24)	Controls (n=24)	Percent change	Significance, p value
ADP	773 HU-cm^2 (0.99)	637 HU-cm^2 (0.99)	+21	<.001
Area	1.05 cm^2 (0.94)	0.87 cm^2 (0.95)	+21	<.002
Radiodensity	746 HU (0.94)	735 HU (0.88)	+1	<.73

ADP, area density product; HU, Hounsfield units.

Note: Mean anterior arch ADP (HU-cm^2), area (cm^2), and radiodensity (HU) in cases and controls, as determined by two raters, with percentage change between cases and controls and statistical significance of that change noted. Interrater correlation coefficient is noted in brackets.

was not specifically assessed. Finally, 4 of the 24 study cases had unilateral or bilateral occipitoatlas fusions in addition to posterior nonunion, features that may potentially alter a patient's biomechanics and independently contribute to the results derived in the study.

Conclusion

It has long been subjectively recognized but not objectively quantified, until the present study, that the anterior arch of the atlas is hypertrophied in cases of posterior arch nonunion. Anterior arch hypertrophy suggests an adaptive osseous remodeling response to chronically elevated mechanical stress and loss of hoop strength in cases of posterior nonunion.

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