Surgical Neurology International

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Case Report

Bipartite atlas in a collegiate football player - Not necessarily a contraindication for return-to-play: A case report and review of the literature

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Received: 04 August 12 Accepted: 22 August 12 Published: 13 October 12

This article may be cited as:
Petraglia AL, Childs SM, Walker CT, Hogg J, Bailes JE, Lively MW. Bipartite Atlas in a collegiate football player - Not necessarily a contraindication for return-to-play: A case report and review of the literature. Surg Neurol Int 2012;3:126.

Available FREE in open access from: http://www.surgicalneurologyint.com/text.asp?2012/3/1/126/102351

Abstract

Background: Congenital malformations of the posterior arch of the atlas are rare, occurring in 4% of the population. Anterior arch aplasia is extremely rare and often only coexists with posterior arch anomalies, resulting in a split or bipartite atlas. This congenital anomaly is believed to be present in only 0.1% of the population.

Case Description: A 19-year-old male collegiate football player presented with neck pain and upper extremity paresthesias after sustaining a tackle that forced neck hyperextension. Computed tomography revealed significant congenital bony anomalies of the cervical spine, with incomplete fusion of the anterior and posterior arches of the atlas; however, there was no evidence for of any acute traumatic injury or fracture. Magnetic resonance imaging revealed increased edema in pre-vertebral soft tissues around C1–C2, with a possible increase in signal within the fibrous ring of the anterior C1 ring. Flexion and extension imaging confirmed reduced range of motion and no instability. Patient was treated non-operatively, and was able to resume normal activity and training regimens, and continued to do well clinically.

Conclusion: We describe a rare case of split or bipartite atlas in collegiate football athlete who sustained a neck injury during a tackle. The patient had no atlanto-axial instability or other clinical contraindications and was managed non-operatively, resuming full participation shortly thereafter with a full resolution of symptoms.

Key Words: Bipartite atlas, cervical spine, neurological sports medicine, return-to-play, spine anomaly

Access this article online

Website: www.surgicalneurologyint.com

DOI: 10.4103/2152-7806.102351

Quick Response Code:

INTRODUCTION

The cranio-vertebral junction is a common site for congenital anomalies and can include malformations of the atlas. Clefts or aplasia of the anterior and posterior arches of the atlas are well documented, but occur rarely.[1,3-8,10,11,14,15,19,21,22,28] The prevalence of congenital malformations of the posterior arch of the atlas can range

from 4 to 5% of the population.[19,21] Anterior arch aplasia is extremely rare, occurring in approximately 0.1% of the population, and often only coexists with posterior arch anomalies.[19,21] Also referred to as "bipartite atlas," the prevalence of combined anterior and posterior defects is uncertain.[11] Cases of combined anterior and posterior clefts of atlas are either asymptomatic or have minimum symptoms, with most found incidentally on imaging studies performed for other indications.[4,10]

Injuries to the cervical spine constitute a common occurrence in those participating in athletic events and can have devastating consequences. These injuries happen primarily to athletes involved in the contact sports of football, wrestling, rugby, and ice hockey, with football injuries constituting the largest number of cases.[2,20] While guidelines for return-to-participation are more evident for those with spinal cord injury or cervical spine instability,[2,12,23-27] there are no clear data to guide the sports medicine practitioner in making such decisions for those with a congenital cleft of the atlas.

In this report, we describe a collegiate football player with a bipartite atlas, which was discovered during the work-up of a neck injury at practice, who ultimately returned to play. We review our patient's case, work-up, and treatment course, as well as the pertinent literature.

CASE REPORT

A 19-year-old male collegiate football player presented to the emergency department (ED) with persistent neck pain following an injury at practice. The contact occurred during a blocking drill and reportedly forced his neck into hyperextension. Upon impact, he immediately experienced high cervical neck pain localized to the left side of his neck, although over time this pain became more generalized. The patient was removed from play, and on the sidelines, no weakness or sensory abnormalities were noted on his examination. The pain was exacerbated with palpation at the base of the skull and did not radiate. Cervical range of motion with rotation, flexion, extension, and lateral flexion was limited secondary to pain. The patient had persistent pain later in the evening, not relieved with analgesics and rest, and was ultimately instructed to go the ED for further work-up and evaluation.

In the ED, he had continued complaint of neck pain; however, he remained neurologically intact and denied numbness, paresthesias, or weakness on initial evaluation. Of note though, later that evening in the ED, the patient transiently experienced subjective numbness in his left lateral forearm in an ulnar distribution that later spontaneously resolved. He was an otherwise healthy individual with a medical history significant for one "stinger" in the past that completely resolved. Plain films revealed no acute fracture or dislocation [Figure 1]. A computed tomography (CT) scan of his cervical spine demonstrated absent bony fusion of the anterior midline synchondrosis, as well as the midline posterior arch of the C1 bony ring [Figure 2a–c]. The osseous components had well-developed cortical margins, strongly suggesting that the midline discontinuities were not the result of trauma. The patient additionally underwent magnetic resonance imaging (MRI) of the cervical spine that demonstrated prominent edema in the pre-vertebral soft tissues extending from the level of the clivus to the vertebral body of C5 [Figure 3a]. Images at the level of the C1 bony ring demonstrated edema of the paramedian ventral soft tissues at, above, and below the level of the unfused anterior midline synchondrosis [Figure 3b and c]. No ligamentous disruption was identified.

The patient's overall clinical picture and imaging findings were consistent with a cervical sprain/strain with an incidental finding of a congenital bipartite atlas. Given his persistent neck pain, tenderness, and findings of pre-vertebral soft tissue swelling, the patient was advised to remain in a rigid cervical collar until seen in follow-up 1 week later. He was treated symptomatically with non-steroidal anti-inflammatory drugs (NSAIDs) and a muscle relaxant. He was temporarily removed from contact drills, but was cleared to ride a stationary bike and to do isometric neck strengthening with the athletic training staff, while remaining in the collar. Follow-up 1 week later revealed diminished tenderness and increased range of motion. Flexion/extension radiographs were obtained and demonstrated no evidence of instability or misalignment. The patient was allowed to resume usual activity and his cardiovascular training regimen with symptomatic restrictions. We opted to wait until his MRI findings resolved, prior to resuming contact activity. Repeat MRI of his cervical spine approximately

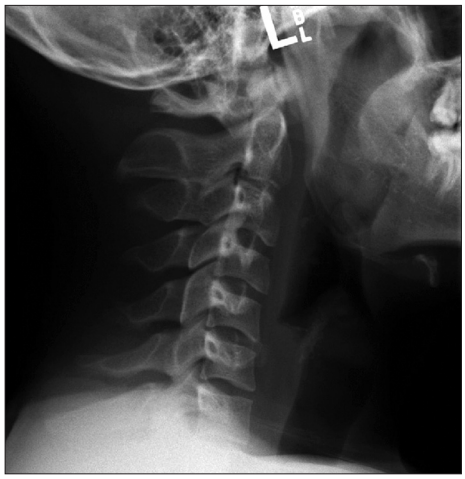

Figure 1: Lateral cervical spine X-ray. Lateral plain film obtained on admission revealed some pre-vertebral soft tissue swelling; however, no acute fracture or dislocation was appreciated

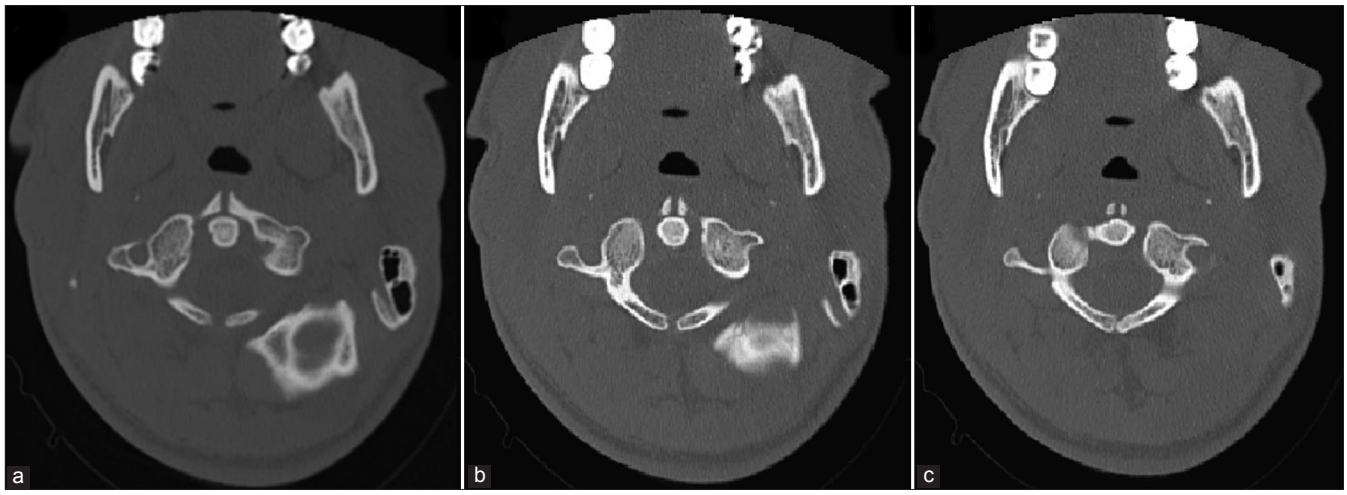

Figure 2: Computed tomography (CT) of the cervical spine. Axial CT images (a–c) demonstrate absent bone fusion of the anterior midline synchondrosis, as well as the posterior midline portions of the C1 bony ring. The osseous components have well-developed cortical margins, strongly suggesting that this midline discontinuity is not the result of trauma

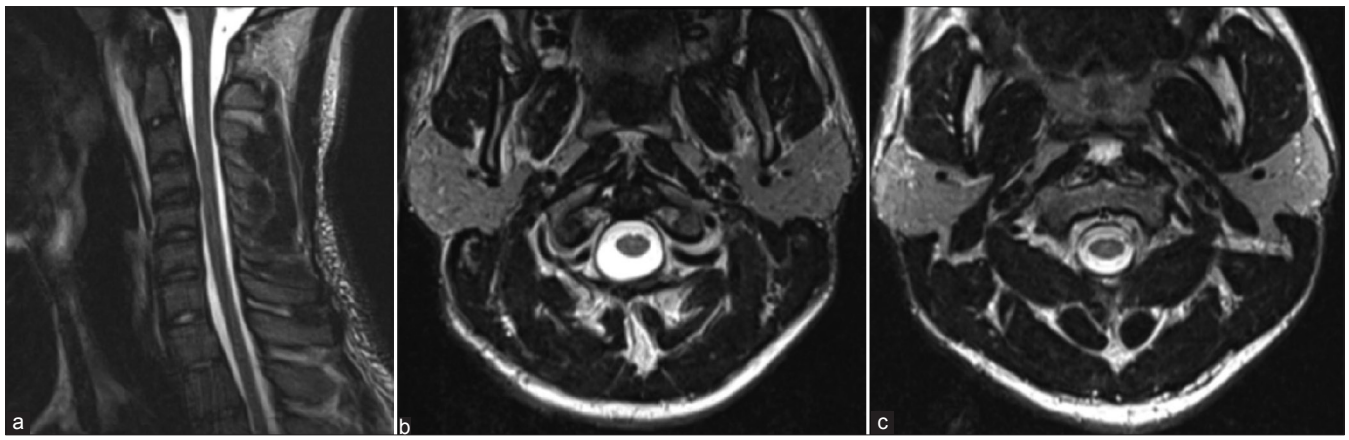

Figure 3: Magnetic resonance imaging (MRI) of the cervical spine. Sagittal (a) T2-weighted MRI image showing prominent pre-vertebral hyperintensity and fusiform swelling of the pre-vertebral soft tissues, extending from the level of clivus to inferior aspect of the C5 vertebral body, indicating edema. No ligamentous disruption is identified. Axial (b and c) T2-weighted MRI images at the level of the C1 bony ring demonstrate the edema of the paramedian ventral soft tissues at, above, and below the level of the unfused anterior midline synchondrosis

1.5 months following the injury demonstrated complete resolution of the pre-vertebral swelling and hyperintensity, and an unchanged appearance of the unfused anterior midline synchondrosis. Even after having a discussion with the patient regarding the uncertain future risk for cervical spine injury and was demonstrated understanding the potential consequences, he desired to continue playing. Given the absence of any signs of instability, the patient was cleared to fully return-to-play. He resumed contact activity, both practice and game-time play, without further issues and continues to remain asymptomatic.

DISCUSSION

Development and incidence

The embryological development of the atlas is complex and various developmental anomalies have been reported. Three ossification centers are responsible for its structural formation: an anterior ossification center which gives rise to the anterior tubercle, and two lateral masses which form the corresponding lateral masses and posterior arch.[5,7,15] A fourth ossification center has been cited on occasion and appears to form the posterior tubercle.[21]

During gestation, the two lateral ossification centers extend posteriorly toward the midline, and form early portions of the posterior arch.[21] Throughout early development, these primitive arches continue to advance, eventually fusing around the fourth year of life.[4] Incomplete fusion of the posterior atlas is estimated to occur in 4% of the population, and is believed to be a result of a failure in chondrogenesis rather than a failure in ossification.[9,14,21] Due to the relative heterogeneity of posterior malformations, a classification scheme was developed by Curarrino *et al*. to categorize them based upon the extent of absence of the posterior tubercle.[6]

Of the five categories, two general types exist: median clefts (Type A) and various degrees of posterior arch dysplasia (Types B–E),[6,28] although the former appears to be present most often.[14,21]

Fusion defects involving the anterior arch of the atlas are much less common, occurring in approximately 0.1% of patients.[3,8,17,28] Numerous studies have estimated anterior arch ossification and synchondrosis fusion to occur somewhat later than for the posterior arch, typically between ages 6 and 8.[16,18] Recent studies, however, have suggested that ossification may take even longer in some individuals.[13,19] Interestingly, unlike anomalies of the posterior arch, anterior clefts rarely occur in isolation and often coexist with posterior arch defects.[10,21] This results in a split, or bipartite atlas,[9,22] as observed in our patient.

Clinical implications

A review of the current literature yielded a limited number of prior bipartite atlas cases reported. In general, these anomalies are considered relatively benign and most cases are found incidentally in asymptomatic patients. A few other reports have been described in the setting of athletic participation.[4,11] One of the reports made no mention of return-to-play recommendations, but noted that the football player was treated symptomatically and ultimately remained asymptomatic.[4] Jans *et al*. chose to recommend that their patient refrain from further participation in contact sports and to "adjust his recreational activities;" however, the authors acknowledged a lack of hard evidence on which this decision was based.[11] Of note, the patient in that case report had no studies demonstrating any evidence of instability.[11]

It is well established that there is a significant risk of cervical spine injury in athletes participating in American football.[20] The rare nature of this cervical anomaly certainly accounts for the lack of literature available to guide management in these situations. The patient lacked any absolute or relative contraindications to return-to-play as designated by accepted treatment guidelines.[2,12,24] Our patient was asymptomatic up until this point in his life, including many years of playing contact sports, and dynamic imaging with flexion–extension radiographs at the time of his cervical strain/sprain revealed no signs of injury or instability. As such, the patient was treated with conservative supportive therapy and we decided to withhold the patient from contact sports until the MRI hyperintensities resolved. The patient was extensively counseled regarding the unclear potential for risk of injury from further participation in football and the uncertainty of whether his risk was any greater than players without his cervical spine anomaly. He demonstrated understanding about the inability to quantify any actual risk and afterward stated unequivocally that he was willing to accept all risks or potential consequences, and wanted to continue in football without restriction. The patient was eventually allowed to return-to-play and participated in both football practice and game-time play without issue.

CONCLUSION

Our report presents an interesting case of an athlete who suffered a cervical sprain/strain and was incidentally found to have a bipartite atlas on subsequent work-up. Most patients with this congenital anomaly are asymptomatic and only rarely are there issues with cervical spine stability. There is little in the way of medical literature to guide management regarding return-to-play in this situation; however, our case provides an example in which the patient was allowed to return to participation in American football. If the player is asymptomatic and there is no evidence of instability on imaging, a bipartite atlas should not necessarily represent a contraindication for return-to-play. Further research is needed to help define the most appropriate management recommendations for patients with congenital atlas anomalies, particularly bipartite atlas.

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