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Bipartite atlas or Jefferson fracture? A case series and literature review

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Study Design

Case series and literature review.

Objective

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We present a new case of a C1 anterior arch fracture following suboccipital craniotomy and C1 laminectomy to add to three previously reported cases, thus increasing awareness of this post-operative complication. We also present two adult cases of presumably congenital bipartite atlas. The radiographic similarity of these cases raises the possibility of a traumatic cause for some instances of bipartite atlas rather than a congenital defect.

Summary of Background Data

The bipartite atlas is a rare entity that has previously been presumed to represent a congenital variant. Its imaging appearance may be mistaken for a Jefferson fracture in the setting of trauma.

Methods

The first case is a 4-year-old female who underwent suboccipital craniotomy and C1 laminectomy and was monitored over the course of the subsequent year. Two cases of bipartite atlas in young males without a history of recent trauma were observed over a similar time period and are presented for comparison.

Results

In the first case, imaging demonstrated the development of an anterior arch fracture following the C1 laminectomy surgery. Correlation of the pediatric and adult cases revealed that it may be difficult to differentiate a subacute or chronic anterior arch fracture from a congenital cleft, leading to the hypothesis that the etiology of the bipartite atlas may be traumatic rather than congenital in some cases.

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Review of the literature revealed that isolated anterior arch defects are extremely rare, and they are more commonly seen in conjunction with posterior arch defects which further supports our hypothesis from a biomechanical perspective.

Conclusion

C1 laminectomy may increase the risk of an anterior C1 arch fracture. A traumatic mechanism is proposed as a cause of the bipartite atlas. In addition, the bipartite atlas may have a higher prevalence than previously reported.

Key Words: C1 fracture, C1 cleft, C1 defect;bipartite atlas, split atlas, anteroposterior spondyloschisis, anteroposterior rachischisis, C1 anterior arch, C1 posterior arch, C1 laminectomy

Level of Evidence: 4

We report a case of a C1 anterior arch fracture following suboccipital craniotomy and C1 laminectomy to add to three previously reported cases. We also present two cases of presumed congenital bipartite atlas. Finally, we postulate a traumatic mechanism for the bipartite atlas rather than a congenital cause.

1. C1 laminectomy increases the risk of a C1 anterior arch fracture.
2. Bipartite atlas is a rare entity with combined defects of the anterior and posterior C1 arches, currently thought to be congenital.

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3. The association between a C1 posterior arch defect and subsequent anterior arch fracture raises the possibility of a traumatic mechanism for the bipartite atlas.

Introduction

Six cases of C1 anterior arch fracture following a suboccipital craniectomy with C1 laminectomy or in association with a posterior arch defect have been reported[1-5]. It has thus been proposed that a C1 laminectomy increases the risk of a C1 anterior arch fracture. We present a case of a proven C1 anterior arch fracture following suboccipital craniotomy and C1 laminectomy in a child, together with two adult cases of bipartite atlas. Based on the similar appearances, we postulate a traumatic mechanism for the etiology of some cases of bipartite atlas.

Case Report

Patient 1 was a 4-year-old female who underwent suboccipital craniotomy with C1 laminectomy for resection of a cerebellar pilocytic astrocytoma. Postoperative head CT demonstrated an intact C1 anterior arch (Figure 1a and 1b). The patient returned to the emergency room 5 months after surgery with neck pain and stiffness for 2 days. CT of the neck demonstrated a mildly displaced fracture of the C1 anterior arch (Figure 2a). Without the prior study for comparison, this fracture could have been mistaken for a congenital cleft of the C1 anterior arch. The patient was treated with a supportive cervical collar for 6 weeks when a CT of the cervical spine revealed widening of the fracture gap (Figure 2b), which resembles a previously reported case of C1 anterior arch fracture in a patient with a congenital posterior arch defect[3]. This case also resembles a reported case of an apparent bipartite atlas where the midline anterior arch defect was later confirmed to be a Jefferson fracture[12].

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Two cases of bipartite atlas were observed in the same month in our adult hospital (Figure 3a and 3b). Patient 2 was a 20-year-old male with a history of seizures. CT demonstrated midline anterior and posterior arch defects. The anterior arch defect was similar in location and orientation to the pediatric fracture described above. However, this defect demonstrated smooth well-corticated margins and was presumed to represent a congenital bipartite atlas. Patient 3 was a 39-year-old male with a history of polysubstance abuse who presented with gait disturbance and dizziness. CT of the head and CT angiogram of the neck revealed a bipartite atlas with a similar appearance to Patient 2. These patients reported no history of recent trauma, supporting the conclusion that the C1 defects were chronic.

Discussion

Congenital anterior arch anomalies are rare and are almost always seen in combination with a posterior arch defect[6,7]. Only 9 cases of isolated anterior arch defects in adult patients have been published (Table 1). The fact that a congenital anterior arch defect is more rare can be partially explained by C1 ossification patterns in that the anterior arch often has one ossification center at the midline whereas the posterior arch commonly has two ossification centers that fuse in the midline posteriorly[8]. However, this reasoning cannot explain why there are more reported cases of combined anterior and posterior defects. It also does not explain why nearly all reported anterior arch defects are in the midline, rather than off-center as would be expected with the most common single anterior ossification center configuration (Figure 4)[17].

We postulate two possible mechanisms for the bipartite atlas: a congenital etiology and a traumatic etiology. The prevailing hypothesis is that there is congenital synchronous or

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metasynchronous failure of osteogenesis or chondrogenesis in multiple ossification centers that form the C1 arch, resulting in both posterior and anterior arch defects during childhood development (Figure 4)[13]. However, we suggest that a congenital posterior arch defect due to failure of fusion of the posterior synchondrosis may subsequently result in a subclinical C1 anterior arch fracture in a small percentage of the population. After healing, this anterior arch fracture develops corticated margins and may be mistaken for a congenital cleft on later radiographic studies. Our case series as well as the six similar prior case reports (see Introduction) would support such a traumatic mechanism. The C1 posterior arch likely plays a pivotal role in distributing axial loading forces, and disruption of this arch may therefore result in increased force on the anterior arch and increased fracture risk.

Conclusion

This case together with three previously reported cases raises the concern for increased risk of anterior C1 fracture following C1 laminectomy. In addition, the association between a posterior arch defect and subsequent anterior arch fracture may help explain the etiology of the so-called bipartite atlas. Anterior arch defects in the setting of a bipartite atlas may represent a fracture consequent to the posterior arch defect rather than a separate congenital cleft.

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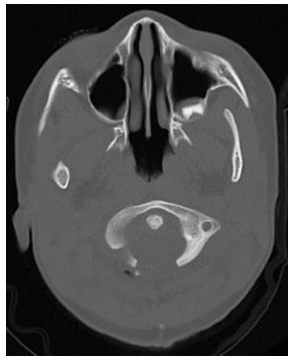

Figure 1a. CT of Patient 1 obtained on postoperative day 1 shows resection of the C1 posterior arch. The anterior arch is intact.

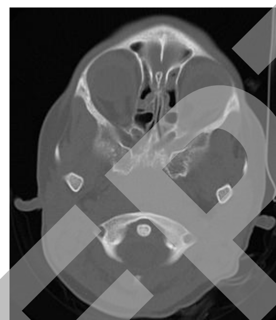

Figure 1b. CT obtained 2 months postoperatively again demonstrates an intact C1 anterior arch.

Figure 2a. CT of Patient 1 obtained 5 months postoperatively now shows a midline well-corticated linear lucency through the C1 anterior arch consistent with a fracture, likely subacute.

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Figure 2b. CT obtained 7 months postoperatively demonstrates a widened fracture gap measuring 5 mm.

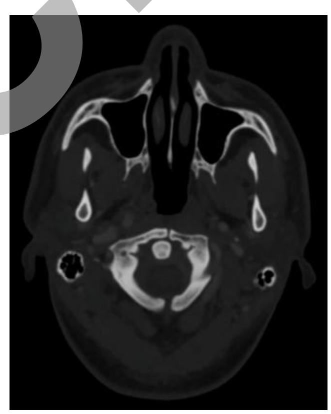

Figure 3a and Figure 3b. Bipartite atlas incidentally noted in Patient 2 and Patient 3 respectively. Also note the prominent notches in the medial aspects of the C1 lateral masses bilaterally.

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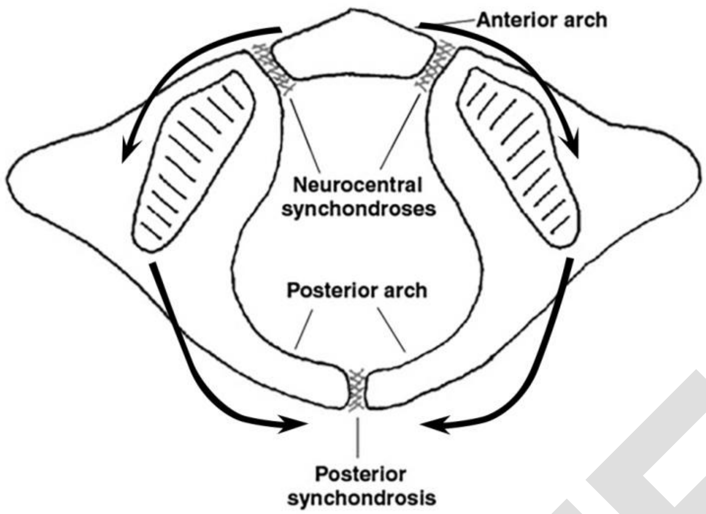

Figure 4. Typical C1 synchondroses, primary ossification centers and direction of ossification. Note that the anterior arch may have 0, 1, 2, 3, or 4 ossification centers. The diagram depicts the most common scenario with a single anterior ossification center and two lateral ossification centers. Reprinted with permission from the Journal of the Canadian Chiropractic Association and the American Journal of Roentgenology[13,17]

Table 1. Tabulation of all cadaveric or CT documented cases of isolated anterior arch defects (i.e. without a documented associated posterior arch defect) in adult patients greater than 18 years of age.

Author	Year	Type	Age	Sex	History
Geipel P[8]	1955	Cadaver	-	-	-
Desgrez H[9]	1965	Cadaver	-	-	-
Mace SE[10]	1986	CT	19	M	Neck pain after fall down 15 steps while intoxicated

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Glasser SA[11]	1991	CT	22	M	Neck pain after unrestrained motor vehicle collision
Chambers AA[12]	1992	CT	18	M	Facial trauma after motor vehicle collision
Van der Velde GM[13]	1997	CT	19	M	Neck pain after football injury
Sasaka KK[14]	2006	CT	75	M	Unresponsive after fall down stairs
Thavarajah D[15]	2012	CT	35	F	Neck pain after go-kart crash
He Q[16]	2012	CT	46	F	Neck pain after fall from 1.5 m height

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