Unilateral atlanto-occipital injury: A case series and detailed radiographic description

ABSTRACT

Context: Atlanto-occipital dissociation is a highly lethal ligamentous injury at the craniocervical junction (CCJ). Previous studies have described rare cases of milder forms of atlanto-occipital injury (AOI) which might be managed nonoperatively, but there is a paucity of literature on this subject.

Aims: We retrospectively reviewed our institutional experience to characterize the injury patterns, treatments, and clinical courses of patients with unilateral AOI.

Methods: We included patients with radiographic evidence of unilateral occipitocervical joint capsular disruption, distraction, or edema ± injury of the apical ligament, tectorial membrane, anterior atlanto-occipital membrane, posterior atlanto-occipital membrane, alar ligaments, or cruciate ligament. The long-term outcomes were gathered from medical records, and six patients were available for Neck Disability Index via phone call at the time of the study.

Results: Eight patients were included in the study. The mean age was 45.1 years ± 26.5. Causes of trauma included motor vehicle collision for five patients (5/8, 62.5%), falls for two (2/8, 25), and assault for one (1/8, 12.5%). All patients had a widened condyle-C1 interval >2 mm. Three patients underwent occipitocervical fusion, one patient underwent atlantoaxial fusion, and another received subaxial fusions for other injuries. Three patients underwent no surgical intervention. All patients were seen at least once as an outpatient following hospital discharge. There were no delayed neurologic injuries or deaths.

Conclusions: We propose that ligamentous injury at the CCJ functions more as a spectrum rather than dichotomous diagnosis, of which a subset can likely be safely managed nonoperatively.

Keywords: Atlanto-occipital dissociation, cervical spine, craniocervical junction, trauma

INTRODUCTION

Atlanto-occipital dissociation (AOD) is a highly lethal ligamentous injury at the craniocervical junction (CCJ) with high risk for the neurologic deficit.[1] Before the recent decades, there was minimal literature on this injury pattern due to the low likelihood of patient survival to tertiary trauma care.[2] However, with improvements in cervical immobilization and prehospital care, more patients are surviving to diagnosis and treatment.[3] As the literature surrounding, AOD has grown, one evolving topic is the clinical and radiographic definition of the injury.[4,5] Because of the, often, subtle nature of the injury and the extremely high risk of neurologic compromise with delayed diagnosis, a variety of radiographic criteria, largely based on plain film and computed tomography (CT), have been established.[4,6]

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However, with the increasing use of magnetic resonance imaging (MRI) in these patients, it is now understood that what was once considered a single diagnosis of AOD is more likely to represent a spectrum of ligamentous injuries that may warrant more nuanced approaches to treatment.[7,8]

In particular, incomplete or unilateral AOD is an entity that has been described in some published series but is often analyzed as part of the broader AOD cohort without separate consideration.[6] Others have also proposed the concept of a milder craniocervical injury within the spectrum of AOD that can be managed more conservatively than the more severe forms;[7,8] however, clinical data regarding this entity are lacking. As a result, the purpose of this study was to utilize our multidisciplinary institutional level 1 trauma center experience to retrospectively characterize and describe the injury patterns and clinical courses of patients with incomplete unilateral occipitocervical injury, which we have termed "Atlanto-occipital injury (AOI)."

METHODS

Patient selection

After obtaining an approval from the Institutional Review Board at the University of Alabama at Birmingham, a case series was retrospectively constructed utilizing the Nuance Primordial Software (Product Information) to search all documented radiology reads from January 1, 2006 to August 1, 2021, for the following phrases and terms: ([edema OR disruption OR avulsion OR tear] AND ["Occipitocervical joint" OR "alar ligament" OR "apical ligament" OR "tectorial membrane" OR "anterior atlanto-occipital membrane" OR "posterior atlanto-occipital membrane" OR "cruciate ligament"]) NOT knee.

A total of 727 results were yielded by this initial search. The radiology interpretations from the queried scans were used to screen potential patients for inclusion by one of the primary investigators Jacob Lepard (JL). Following the initial screening process, the CT cervical spine and MRI cervical spine images were directly evaluated for study inclusion. Patients were included, if they were older than 18 years with a history of recent traumatic injury and radiographic evidence of unilateral occipitocervical joint capsular disruption, distraction, or edema ± injury of the following structures: apical ligament, tectorial membrane, anterior atlanto-occipital membrane, posterior atlanto-occipital membrane, either of the alar ligaments or transverse segment of the cruciate ligament. All patients with evidence of contralateral occipitocervical joint injury were excluded from the study. In addition, patients were excluded, if there was evidence of nontraumatic craniocervical pathology like

infection or chronic degeneration. Demographic data and clinical outcomes were retrospectively obtained by chart review and summarized using descriptive statistics. Phone contact was attempted for all patients at the time of the study for updated assessment of their disability status due to their cervical injury. For those patients in whom contact was successful, Oswestry Disability Index, Neck Disability Index (NDI), and pain Visual Analog Scales (VAS) were obtained.[9] Injury severity scores (ISS) were obtained from a prospectively collected trauma database with a range of 1–75 and a score >15 indicating a major traumatic injury.[10]

Radiographic measurement

At our tertiary care level 1 trauma center, all patients undergo Advanced Trauma Life Support protocol after traumatic injury with a standardized imaging protocol. Using CT imaging of the cervical spine, the following parameters were measured using previously published methods: revised condyle-C1 intervals (CCI), Powers ratio, basion-axial intervals (BAI), and basion-dental intervals (BDI).[1] Measurements were considered to be abnormal, if the CCI was >2 mm, Powers ratio >1, BDI >10 mm, or BAI <−4 mm or >12 mm.[11] In addition, cranial CT was utilized to evaluate for cervicomedullary traumatic subarachnoid hemorrhage, and when available, CT angiogram of the neck was used to identify associated blunt cervical vascular injury. The occipitocervical joints, ligamentous structures, and the CCJ were evaluated using MRI cervical spine. Prevertebral soft-tissue swelling was measured on the midsagittal CT at the level of the C2 endplate with thickness >6 mm representing abnormal edema.[12] In one patient (Patient 7), CT imaging of the cervical spine was not available, and so all radiographic parameters were measured using MRI only. No patients underwent cervical traction testing.

RESULTS

Demographics

A total of eight patients were included in the study. There were six (6/8, 75%) male and two (2/8, 25%) female. The mean age was 45.1 years ± 26.5 and with a range of 19–91. There were six Caucasians (6/8, 75%) and two African–Americans (2/2, 25%). Causes of trauma included motor vehicle collision for five patients (5/8, 62.5%), falls for two (2/8, 25), and assault for one (1/8, 12.5%). Five patients (5/8, 62.5%) had an American Society of Anesthesiologists score of 3, two patients (2/8, 25%) had a score of 4, and one patient's score was not documented. ISS were available for eight patients. The average ISS for the cohort was 18.9 ± 10.9 with a range of 10–41 [Table 1].

Radiographic findings

On CT imaging, five patients had a right-sided sagittal CCI, and three patients had a left CCI that was >2 mm. One

patient had a BDI >10 mm. No patients were found to have an abnormal BAI or Powers ratio. Significant prevertebral edema was present in three patients (3/8, 37.5%). One patient had noncompressive retroclival subdural hemorrhage, and there were no patients with cervicomedullary subarachnoid hemorrhage. One patient had a unilateral intimal injury of their vertebral artery at the site of a coincident subaxial injury. In the remaining seven patients, there was no evidence of vertebral artery injuries. Three patients had concomitant occipital condyle fractures, one patient had a Jefferson fracture, two had evidence of ligamentous injury at C1-2, two patients had cervical 2 fractures, and two patients had subaxial cervical spine injuries [Table 2].

MRI was performed on all included patients. Indications for MRI included evaluating injury at C2 or above in five patients, evaluating subaxial injuries in one patient, and cervical collar clearance in two patients that were intubated and sedated. Six patients had increased short tau inversion recovery (STIR) signal in an occipitocervical joint unilaterally that corresponded to increased CCI on CT imaging [Figure 1]. Two of the eight patients with unilaterally increased CCI did not have a corresponding STIR signal on MRI. Three patients had unilateral and three had bilateral alar ligamentous injury. Only one patient demonstrated injury to the atlanto-occipital

membrane, while three had injury to the posterior atlanto-occipital membrane. One patient had complete and another had partial injury to the tectorial membrane. One patient had a complete rupture of the ascending portion of the cruciate ligament, and another four had injury to the transverse ligaments. Two patients were found to have a complete disruption; another had a partial tear to the apical ligament. Finally, seven patients had increased STIR signal change in the interspinous ligaments and soft tissues between the occiput and cervical 2 [Table 3].

Treatment

On presentation, six patients were Glasgow Coma Scale (GCS) 15 and neurologically intact and another two patients were GCS 10T without evidence of deficits on the neurological examination. Three patients underwent occipitocervical fusion, one patient underwent atlantoaxial fusion, and another received subaxial fusions for other injuries. Three patients underwent no surgical intervention and were treated with Aspen collar only. The average hospital stay was 11.3 ± 11.1 days with a range of 5–36 days. Four patients were discharged directly to the home, two patients were discharged to inpatient rehab, one required skilled nursing facility care, and one died of their traumatic injuries. There were no delayed complications or late-onset neurologic deficits in this cohort. All patients were seen at least once as an outpatient following hospital discharge. The average follow-up was 26.4 ± 45.1 months with a median of 10.7 months and a range of 0.9–126 months [Table 4].

Clinical outcomes

Three patients were available for a phone follow-up at the time of data collection. Of these, all three complained of ongoing neck pain. The mean NDI score was 33.5 ± 6.06 and the mean VAS was 4.3 ± 1.5 at a mean interval of 55.4 months from injury. One of the four patients underwent occipitocervical fixation, whose NDI was 28% and VAS was 4. The remaining two patients did not undergo occipitocervical fixation and had a mean NDI score of 36.3 ± 5.3. Based on

Table 1: Demographics and baseline clinical information

Patient	Gender	Age at injury	Ethnicity	Mechanism of injury	ASA	Injury severity score
1	Male	66	White	MVC	3	13
2	Male	56	White	Fall	3	10
3	Female	20	AA	MVC	4	22
4	Male	91	White	Fall	4	10
5	Male	28	AA	MVC	3	14
6	Female	19	White	MVC	3	41
7	Male	23	White	Assault	-	-
8	Male	58	White	MVC	3	22

ASA - American Society of Anesthesiology; AA – African–American; MVC - Motor vehicle collision

Table 2: Computed tomography characteristics

Patient	Right CCI	Left CCI	BDI	BAI	Powers ratio	Prevertebral edema (mm)	SAH	Vertebral artery injury?	Additional cervical injury
1	3.3	1.3	4.7	−2.7	0.67	No	No	Intimal Injury	Right type III OC fracture, C7 pillar fracture
2	1	2.9	8.4	6.6	0.8	No	No	No	Left type II OC fracture
3	4.5	1.9	11.3	6.1	0.81	15	No	No	C1-2 ligamentous injury
4	3.9	1.2	7.8	2	0.78	No	No	No	Type II odontoid fx, C1-2 joint widening
5	3.4	1.2	10	6.5	0.77	7.7	No	No	Right type II OC fracture, Left type III OC fracture
6	1.4	2.1	5.4	2	0.65	No	Clival SDH	No	C2 lateral mass fracture
7	1.9[†]	2.9[†]	3[†]	5.9[†]	0.94[†]	6	No	No[†]	Jefferson fracture
8	3.6	1.8	10	9.8	0.95	No	No	No	C6-7 extension injury

[†]No CT available. Measurements made based on MRI. CCI - Condyle-C1 interval; BDI - Basion-dental interval; BAI - Basion-axial interval; SAH - Subarachnoid hemorrhage; SDH - Subdural hemorrhage; OC - Occipital condyle; MRI - Magnetic resonance imaging; CT - Computed tomography

Table 3: Magnetic resonance imaging characteristics of included patients

Patient	Reason for MRI	Right OC STIR	Left OC STIR	Alar	Ant-AOM	Post-AOM	Tect Memb	Cruc Lig	TAL	Apical	Interspinous
1	Evaluate C7 fracture	Yes	No	No	No	No	No	No	No	No	Yes
2	Clear c-collar, intubated	No	Yes	Right only	No	Yes	No	No	P	No	Yes
3	OC and C1-2 widening	Yes	No	Bilateral	No	No	P	No	No	Yes	Yes
4	Evaluate odontoid injury	Yes	No	Right only	No	No	No	No	Right only	No	No
5	Evaluate craniocervical junction	Yes	No	No†	No	No	No	No	No	P	Yes
6	C-collar clearance	No	No	Right only	Yes	Yes	Yes	Yes	No	P	Yes
7	Evaluate OC and C1-2 injury	Mild	Yes	Bilateral	No	Yes	No	No	Bilateral	Yes	Yes
8	Evaluate OC injury	No	No	Left only†	No	No	No	No	No	No	Yes

†Associated condyle avulsion. OC - Occipitocervical; STIR - Short tau inversion recovery; Ant-AOM - Anterior atlanto-occipital membrane; Post-AOM - Posterior atlanto-occipital membrane; Cruc Lig - Cruciate ligament; TAL - Transverse atlantalligament; Tect Memb - Tectorial membrane; MRI - Magnetic resonance imaging; P - Partial injury

Table 4: Clinical course and outcomes

Patient	GCS	Weakness?	Surgery	Length of stay (days)	Discharge disposition	Delayed neurologic complication	Follow-up (months)
1	15	No	ACDF C6-7	7	Rehab	No	0.9
2	15	No		11	SNF	No	10.7
3	15	No	O-C3 PSIF		Home	No	27.7
4	15	No		5	Deceased	No	Died
5	15	No	O-C2 PSIF	6	Home	No	126.5
6	E4VtM5	No		8	Rehab	No	1.7
7	15	No	C1-2 PSIF	6	Home	No	4.9
8	E3VtM6	No	O-C2 PSIF	36	Home	No	12.6

GCS - Glasgow Coma Scale; PSIF - Posterior spinal instrumentation and fusion; SNF - Skilled nursing facility; ACDF - Anterior cervical discectomy and fusion

the NDI disability scale, the patient who underwent occipital condyle fusion was rated as mild whereas of the three patients who did not require surgical fixation, two had a disability rating of moderate.

DISCUSSION

It is notable that before approximately 20 years ago, very few published series on AOD existed in the literature. As our understanding of this injury pattern and the biomechanics of the CCJ has grown, it is worth considering the possibility that injury to this mechanically complex region could represent a spectrum rather than binary classification of AOD versus not. In this case series, we have sought to emphasize this point by describing the radiographic findings and clinical course of eight patients with unilateral craniocervical injury that does not represent a complete AOD.

Horn *et al*. acknowledged the presence of these indeterminate cases in their institutional series of 33 survivors of AOD, in which five (5/33, 15.2%) were determined to be a Grade 1 injury, indicating normal CT findings based on the current diagnostic parameters (Powers ratio, BDI, and BAI), but with "moderately abnormal" MRI findings. All five of these patients were managed with external orthoses without late neurologic deterioration.[7] Similarly, Bellabarba *et al*. proposed a three-stage classification of AOD, in which
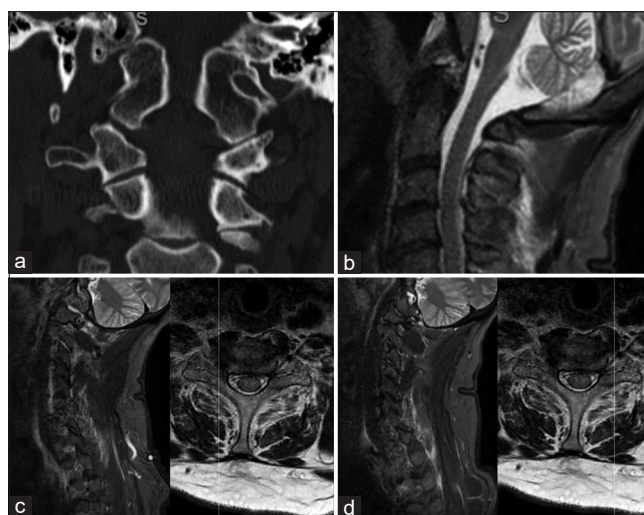

Figure 1: A 66-year-old male presenting after a motor vehicle collision with (a) coronal CT cervical spine, (b) midsagittal MRI, (c) right parasagittal MRI, and (d) left parasagittal MRI demonstrating asymmetric widening of the right condyle-C1 interval with associated STIR signal indicating mild atlanto-occipital injury. He underwent surgery for additional subaxial trauma, but required no fixation for craniocervical injuries. CT - Computed tomography, MRI - Magnetic resonance imaging, STIR - Short tau inversion recovery

Stage 1 patients were defined as "stable minimally or nondisplaced craniocervical injury, in which there is a sufficient preservation of ligamentous integrity to allow for nonoperative treatment." Radiographically, these Stage 1 patients were defined as MRI evidence of ligamentous craniocervical injury with normal alignment on static

radiographs and <2 mm distraction on provocative traction testing.[8] Because these Stage 1 patients were considered, by definition, to not be true AOD, Bellabarba *et al*. did not include them in their series of 17 survivors of AOD, and thus, their presentation and clinical course are not known. In our own series of eight patients with this injury pattern, the most presented after high-velocity motor vehicle crash or falls. On presentation, the vast majority were neurologically intact without lower cranial nerve deficit.

Despite the, often, dramatic instability exhibited by patients with a complete AOD, a unified diagnostic criterion remains elusive.[5] Instead of this, multiple radiographic screening measures have been devised, positive findings in any of which result in concern for AOD and consideration of soft-tissue imaging.[11] In particular, the revised CCI, which was originally proposed by Pang *et al*.,[13,14] has been found to be highly sensitive in the diagnosis of AOD compared to other measures such as the Powers ratio, BAI, and BDI. The findings of our study are consistent with this, as all patients had an abnormal CCI unilaterally, which nearly always correlated with increased STIR signal on MRI. In contrast, only one patient had a mildly elevated BDI, and there were no abnormal BAI's or Powers ratios. Additional radiographic clues for craniocervical injury such as prevertebral swelling, vertebral artery injury, and cervicomedullary subarachnoid hemorrhage were not reliably present in our cohort. Given the definition for abnormal CT used by Horn *et al*., which did not include CCI, seven of our eight patients would have been considered a Horn Grade 1, while the single patient with an elevated BDI would perhaps have been a Grade 2. This highlights the importance of the CCI as a clinically important screening tool that can diagnose the mildest versions of AOI, in which there is clinically important ligamentous injury in the absence of gross instability. It also indicates that the common injury pattern in our cohort is likely consistent with the five Grade 1 patients managed nonoperatively by Horn *et al*.

This consideration of partial atlanto-occipital injuries begs the important question as to what degree of ligamentous injury can be tolerated and at which point the CCJ becomes unstable.

Biomechanical studies have attempted to elucidate the most important structures for the maintenance of stability at the CCJ with conflicting results. Phuntsok *et al*. performed a finite element analysis of the upper cervical ligaments finding that the greatest decrease in stability occurred with injury to the occipitoatlantal capsular ligaments.[15] In a cadaveric study, Radcliff *et al*. found the transverse and alar ligaments to be the primary stabilizers at the CCJ.[4] In their review

of the craniocervical ligaments, Tubbs *et al*. noted that there is an ongoing controversy regarding the importance of multiple structures including the apical ligament and tectorial membrane.[16] The most likely synthesis of these studies is that multiple of these ligaments play important roles in the biomechanical integrity of the CCJ to a greater and lesser degree depending on the type of craniocervical motion being considered. While our own series lacks the power to definitively answer the question of what degree of ligamentous injury can be safely managed nonoperatively; it is noteworthy that only three of the eight patients with AOI in our series underwent occipitocervical fixation, while the remainder were managed with external orthosis. By definition, all patients in our series had unilateral injury to the occipitoatlantal capsular ligaments with varying degrees of integrity of the midline ligamentous structures. Despite all having similar injury patterns, there were no late neurologic deteriorations or missed instability in the nonoperative group compared with the operative. When contacted by phone, with a mean follow-up of 55 months from injury, the mean NDI score among the two nonoperative patients was higher than the one patient who underwent occipitocervical fixation. While this is a trend worth investigating, the low numbers available for phone contact preclude a solid conclusion regarding the impact of operative fixation versus conservative management on long-term pain scores.

Our study is limited by the retrospective nature of the data collection. In addition, while loss to follow-up is well-documented in the trauma population,[17] it remains a limitation in the phone follow-up component of our study such that less than half of the included patients were successfully contacted. Furthermore, because all treatment decisions were made by the consulting surgeon at the time of injury, it cannot be definitively known whether the same criteria for nonoperative versus operative management were used for all patients. Further investigation would benefit from a prospective cohort methodology, in which consistent operative criteria were used. Lastly, in focusing our search only to patients with unilateral injury we have likely introduced selection bias to the radiographic characteristics of our cohort. There are likely other anatomic varieties of incomplete AOD or AOI, which are not strictly unilateral, that might also have been included in this study. However, in attempting to demonstrate the concept of incomplete AOD, we sought to create as homogenous a cohort as possible regarding injury patterns. Future studies might include a multi-institutional collaboration to seek out larger numbers of these patients with incomplete AOD, from which better definitions and radiographic criteria might be derived.

CONCLUSIONS

We report here a series of eight patients who sustained atlanto-occipital injury as evidenced by unilateral injury of their occipitoatlantal capsular ligaments. While three underwent occipitocervical fixation, the remaining five required only external orthosis for their craniocervical injury. This raises the important consideration as, to whether injury to the biomechanically complex CCJ can be adequately simplified to a binary classification such as the presence or absence of AOD. We propose instead that ligamentous injury in this region is more accurately considered a spectrum, of which a subset can likely be safely managed nonoperatively. Further investigation will be required to adequately define safe operative criteria.

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Conflicts of interest

There are no conflicts of interest.

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