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Atlantoaxial Rotatory Subluxation in Children

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Abstract

Objectives—Pediatric cervical injuries are uncommon. This study was to describe injury circumstances, clinical findings, and management among children diagnosed with atlantoaxial rotatory subluxation (AARS) to aid in its recognition and management.

Methods—Subanalysis of a large case-control study January 2000 to December 2004 in seventeen hospitals in the Pediatric Emergency Care Applied Research Network. Cases were children younger than 16-years-old with AARS after blunt trauma (n=55); controls were a.) children with other cervical spine injuries (other CSI) (n=485) and b.) those with normal imaging of the cervical spine (non-CSI) (n=1060).

Results—Children with AARS were younger: (mean (SD) age 7.7 (3.8) vs. 10.7 (4.6), Wilcoxon $p<0.01$). Falls accounted for 36% of injuries; there were no diving mechanisms (vs. other CSI, falls 19%, Fisher's exact $p<0.01$; diving 7%, $p=0.04$). Children with AARS sought medical care more than 24 hours after the injury event (21% vs. 1% for non-CSI controls, $p<.01$). Clinical findings associated with AARS included neck pain (67%) and torticollis (57%) vs. other CSI, pain (47%) torticollis (5%) $p\leq.01$ for each, and vs. non-CSI controls, pain (33%) and torticollis (6%), $p<.01$ for each. Management of AARS included no intervention (n=6, 11%), soft or rigid collar only (n=24, 44%), traction (n=14, 25%), halo (n=9, 16%), and internal fixation (n=2, 4%) and varied across institutions ($p=0.02$).

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Conclusions—Children with AARS often have a delayed presentation with neck pain and torticollis; falls are a common injury mechanism. Treatment varied across institutions. Further work is needed to identify optimal management.

Keywords

neck injuries; cervical; spinal injuries

Introduction

Children with trauma involving the head or neck may describe neck pain or stiffness, report bony or muscular tenderness, or hold the head in a tilted position (torticollis). Cervical spine injuries (CSI) associated with blunt trauma are rare events, and minor, nonspecific muscle strain is the most common diagnosis when evaluating for potential CSI.[1,2] Atlantoaxial rotatory subluxation (AARS), also called traumatic torticollis, is one of the more common cervical spine injury patterns in children; with rotation, the lateral masses of the first cervical vertebrae sublux relative to the second cervical vertebrae. Young children have lax ligaments and robust synovium that allows this to occur, as a result AARS occurs almost exclusively in children. AARS may be associated with trauma, infection of the pharyngeal space (Grisel's Syndrome), or occur with no precipitating event.[3,4] Acutely, the patient assumes a "cock-robin" position with the head rotated to one side and the neck tilted laterally toward the opposite side. Over time, the neck pain decreases, and there is a chronic loss of cervical rotation. There is variability in management, which appears in part to be based on duration and severity of signs and symptoms. Accurate, timely diagnosis is important as children presenting within 3 weeks of symptom onset and identified are often managed conservatively (cervical collar or traction). Those who present and are recognized after longer periods of symptoms are more often managed surgically.[5]

Detailed information about the clinical presentation, diagnostic evaluation, treatment and outcome of children with AARS is needed to better understand this disease and standardize care. Pediatric AARS has only been described in case reports and small case series.[5–9] These studies lack defined comparison groups which precludes detailed comparisons about clinical and mechanism of injury factors that would help to distinguish patients at risk for AARS from other diagnoses among children with blunt trauma to the head and neck.

The purpose of this study is to describe injury circumstances and clinical findings to help distinguish AARS from other CSI as well as from children with suspected CSI after blunt trauma that was ruled out (non-CSI controls). We also report management and outcome of children with AARS and compare them to children with other CSI.

Methods

This is a secondary analysis of a previously reported large case-control analysis of children younger than 16 years old presenting to 17 hospitals in the Pediatric Emergency Care Applied Research Network (PECARN) from January 2000 through December 2004 for care of blunt trauma-related injury.[10] In the original study, children with CSI were identified using ICD-9 codes for injury to the cervical spinal cord, vertebrae and/or ligaments, and the medical record was reviewed by the site's principal investigator. The study's principal investigator (XXX) and a collaborating pediatric neurosurgeon (XXX) reviewed all imaging reports and spine consultations to verify the presence of a CSI and assign a CSI classification. For each case of CSI, up to two controls were randomly selected from children who presented for care at the same participant hospital during the same time period as the cases of CSI and were evaluated for blunt trauma with imaging of the cervical spine, but found to be free of CSI. For this analysis, we focused on children with isolated AARS. From the data collected for the original study, we defined two comparison groups: one included all children with other CSI and the second a control group of children with blunt trauma and normal cervical imaging.

Data abstracted from the medical records at each site included use of emergency medical services (EMS), demographic characteristics (age, gender, race), preexisting conditions, injury mechanisms, clinical presentation at the study site, CSI characteristics, other injuries, clinical management, and outcomes at discharge. For motor vehicle crash occupants, we defined as high risk death in the same collision, ejected, head-on collision, rollover, or speed ≥ 55 miles per hour.

The Institutional Review Board at each site approved the study. Abstracted study data were de-identified and stored in secure encrypted databases.

We describe demographic characteristics, injury mechanisms, and clinical findings of the AARS, other CSI, and non-CSI controls using frequencies and percents. We also compare AARS to other CSI in terms of imaging performed, interventions performed, and outcomes. We used Fisher's exact test of homogeneity to compare categorical distributions, and Mann-Whitney-Wilcoxon tests to compare age between cohorts. We used Fisher's exact tests to make other comparisons, including associations between management of AARS and participating institution, highest level of injury, and presence of spinal cord injury, as well as clothes-lining and axial load mechanisms between cohorts. Monte Carlo simulations were used to compute Fisher's exact p-values when direct computation was not feasible. Results were deemed significant if the p-value<0.05. All analyses were performed using SAS/STAT software (Version 9.3, SAS Institute, Cary, NC).

Results

We identified 540 children with CSI; 55 children with AARS and 485 with other CSI. For the children with CSI, we identified 1060 non-CSI controls with blunt trauma and normal cervical imaging. Table 1 reports demographics and clinical presentation. Children with AARS were significantly younger than those with other cervical spine injuries (7.7 vs. 10.7 years). The age distribution was significantly different among the three groups: children younger than 8 years old accounted for more than half of those with AARS while children older than 8 year old accounted for the majority of other CSI. There were similar numbers of boys and girls with AARS; those with other CSI and non-CSI controls both had higher proportions of boys. Children with AARS were less likely to be transported from the scene of injury by EMS and more likely to be transferred from an outside hospital, and/or to have sought medical care greater than 24 hours after the injury event.

The mechanisms of injury are shown in Table 2. For pediatric patients with AARS, falls accounted for 36% of the injury mechanisms, and no child with AARS reported a diving mechanism. Among children with other CSI injuries, falls were less often the injury mechanism (19%); diving accounted for 7% of the injuries. Motor vehicle crash occupant (MVC) mechanism accounted for 11% of injuries among children with AARS, 30% in those with other CSI, and 25% of non-CSI controls. Of the 6 children with MVC-related AARS, 5 were involved in a high risk MVC as defined in the methods.

Among specific injury biomechanics, we observed clothes-lining to be involved in the mechanism for 4 (8%) of those with AARS (vs. other CSI, 2%, Fisher's exact test p=.02, vs. non-CSI controls, 1%, p<.01). Axial load was no different from non-CSI controls, and less than observed with other CSI (AARS 16%, vs. non-CSI controls 22% p=0.40, vs. other CSI 33% p=0.01).

The clinical findings of neck pain and tenderness were common for children in all three groups (Table 3). More than half of children with AARS had torticollis, a substantially higher proportion than those in either comparison group. Children with AARS were less likely than children with other CSI to present with findings of head injury. Focal neurologic findings were most common in children with other CSI (36%); however, they were reported for both AARS (14%) and non-CSI controls (6%). Rates of reported focal findings (paresthesias, sensory loss, motor weakness, and other neurologic findings) were somewhat elevated in those with AARS compared to non-CSI controls. One child with AARS had a condition that predisposed to CSI (achondrodysplasia). Ten of those with other CSI had predisposing conditions (achondrodysplasia n=1, Arnold Chiari Malformation n=4, history of CSI n=1, cervical spinal stenosis n=2, congenital anomaly of cervical vertebrae n=1, Down Syndrome n=1).

Table 4 presents the cervical imaging used in evaluating children with CSI as well as interventions. Children with AARS were less likely than children with other CSI to be evaluated with plain radiographs and magnetic resonance imaging (MRI) and more likely to be evaluated with computed tomography (CT). Across sites, CT use varied. Among the 16 sites with AARS patients, 13 evaluated all AARS patients with CT, two sites evaluated 2 out of 3 patients with CT, and one site did not use CT to evaluate their single patient. Among those with other CSI, CT rates ranged from 26% to 92% across sites. For AARS patients, MRI use varied across sites with 3 sites using MRI for evaluation of all AARS patients, and 7 sites using MRI for none. Among those with other CSI, MRI rates ranged from 37% to 75% across sites.

Diagnosis and management of AARS was site specific. The proportion of youth with CSI diagnosed with AARS differed across sites (Fisher's exact test p=0.01). The site with the highest rate of AARS diagnosed 10 AARS cases out of 22 cases of CSI while another site had no cases of AARS in 18 CSI patients. In addition, interventions for children with AARS also varied widely. Overall, six children with AARS (11%) were managed without intervention. The remainder was evenly divided between management with cervical collars alone (n=24, 44%) and surgical intervention (n=25, 45%). Surgical interventions included internal fixation (n= 2), halo placement (n=9), and traction (n=14). Rates of management techniques for AARS were not consistent across sites (Fisher's exact test p=0.02). Of the 16 sites with AARS patients, 5 (31%) did not use surgical interventions (site n ranging from 1 to 4 per site). The site with the highest rate of AARS (10 patients) managed 3 with traction, and the remaining 7 with cervical collars only. Management by the remaining sites (n ranging from 1 to 6 per site) varied from 40% to 100% surgical management of AARS patients.

Management of AARS differed from management of other CSI (Fisher's exact test p<0.01). None of the AARS were managed with a brace compared to 5% of other CSI. Traction was used as the highest level of management for 25% of AARS, only 4% percent of other CSI were managed with traction; and all additionally received either a halo or internal fixation. Internal fixation for AARS was rare (4%) compared to other CSI (17%).

Outcomes for this study were classified as normal, persistent neurologic deficit, and death, and differences between groups were significant. (Table 5) Among children with AARS, there were 6 (11%) with persistent deficit and no deaths. All cases with poor outcomes had a high-risk mechanism and other substantial injuries (head/torso trauma). There was no association between highest level of intervention and outcome.

Discussion

In this large, multi-center cohort of children with blunt trauma-related CSI, AARS accounted for 10% of injuries. Children with AARS were younger than those with other CSI, often presented greater than 24 hours after injury and with torticollis, and had significant management variation across sites. Cervical spine injuries among children are rare, and to-date AARS has been reported in the literature as single cases or single institution case series.[5–9] The data we report allow us to more comprehensively describe the epidemiology as well as the associated injury mechanisms, clinical findings, and management variation in a larger cohort of children with blunt cervical trauma. This information will help the clinician properly identify children with AARS. We are also able to compare mechanisms and findings to children with other CSI and to children with blunt trauma but without CSI.

Our data are consistent with other studies and indicate that AARS is more often diagnosed among younger children. [6,7,9] In children younger than 8 years of age, ligaments and joint capsules are elastic which allows for hypermobility. The facet joints of C1 and C2 are horizontally aligned and have a robust synovium, which also predisposes to hypermobility. Paraspinal muscles are less well developed in younger children. [11–13] While non-AARS CSI are more common in boys, the gender distribution we observed in those diagnosed with AARS was equally divided between boys and girls. The higher rate of cervical injury among males as compared to females is often attributed to greater exposure to high risk injury mechanisms among males; the data for AARS imply similar exposure for boys and girls to the more minor injury events that are associated with this finding.

The time to presentation after the injury was longer than 24 hours for 20% of children with AARS. This delay is consistent with the reported literature.[5–7] Subach, et al, reported a range of 1–150 days between symptom onset and presentation.[8] Delay in seeking care may occur because AARS can result from rather minor trauma and can be confused with muscle strain. Only 10% of patients with other cervical injuries had a delayed presentation. It is important that primary care providers include AARS in the differential for children who present for care several days after injury and who have characteristic symptoms. A majority of children with AARS presented with torticollis in addition to neck pain and tenderness, whereas torticollis was infrequent in children with other CSI and non-CSI controls.

The physiology of AARS involves subluxation and/or fixation of the atlantoaxial joint in a malrotated position.[3] The main function of the atlantoaxial joint is rotation rather than flexion or extension. A published classification system (Type I-IV) is based on the degree of displacement and integrity of the transverse ligament.[4] This classification system was not routinely reported in the records of children with AARS and as a result, we could not correlate severity of AARS with presentation, interventions or outcome.

As ligamentous laxity appears to be important to the development of AARS, we specifically abstracted information about predisposing conditions including juvenile rheumatoid arthritis, cervical stenosis, Down syndrome, Klippel Feil Syndrome, mucopolysaccharidosis, Ehlers-Danlos Syndrome, Marfan syndrome, achondrodysplasia, and osteogenesis imperfecta. Overall the number of children with these conditions in the data we used was small. Only one child with AARS had achondrodysplasia.

The reported injury mechanism for AARS was most often a fall or other blunt injury to the head or neck. This is consistent with data suggesting minor injury events to be associated with this finding.[5–9] However, more serious injury mechanisms were also represented in our data. We observed no diving or hanging injuries among children with AARS. These data imply that the injury mechanisms associated with AARS are variable: AARS should be included in the differential diagnosis for both minor and more significant mechanisms of blunt injuries to the head or neck.

Some of the children with AARS in our cohort presented with focal neurologic findings and altered mental status. Case series data suggest while focal neurologic findings are infrequent, they have been reported with AARS and associated cord compression [5–9, 13] Of the children with AARS and persistent neurologic deficits, 4 of 6 had a high-risk injury mechanism, and 4 of 6 had a spinal cord injury. The 2 without spinal cord injury had associated substantial head injury. It is likely that both spinal cord injury and associated head injury contributed to the neurologic findings.

Nearly all children diagnosed with AARS had CT imaging. While concern about radiation exposure has led clinicians to question use of CT for general cervical spine imaging, it remains the modality of choice for AARS. Cervical CT, both dynamic CT and CT with three dimensional reconstruction, is currently considered the imaging study of choice.[14–16] It can be difficult to distinguish positional rotation from AARS on plain radiography alone. MRI avoids radiation and provides detail about ligamentous injury and spinal cord trauma, but does not allow for reconstruction or dynamic imaging which is necessary in the diagnosis of AARS.[16] As AARS frequently spontaneously resolves, one option for evaluation of the neurologically intact child with a history and presentation consistent with AARS is to obtain screening plain radiographs and defer CT and the associated radiation exposure until after a failed trial of cervical collar combined with an anti-inflammatory, analgesic and/or muscle relaxant. [5,8]

Management included both hard and soft cervical collars, traction, halo and internal fixation. We did not collect specific information about manual reductions, but one may infer that this was performed before internal fixation and/or halo placement. One-third of children with AARS were placed in traction. Other series have reported higher rates of traction use in AARS patients. [5,8,9] Our data imply management for AARS mostly occurs in the non-operative setting; this has been reported recently by others.[5] Internal fixation is used in those patients with neurological deficits or presenting in a subacute fashion. Significant institutional and surgeon variation exists in the use of collars, traction, and halo immobilization preventing any meaningful conclusions regarding non-operative management. This is possibly related to institutional culture and surgeon background and training. In addition, the duration of symptoms (longer than 24 hours) prior to decision for non-operative or operative management was not recorded, a variable important to future prospective studies.[5]

Limitations

The limitations of this study are inherent to the retrospective methodology and include ascertainment bias and missing data. Although this multicenter effort provided instructions and training relevant to data abstraction, there may have been variability among clinicians about the ascertainment of clinical information documented in the medical record. We used remote and onsite monitoring as well as inter-rater reliability measurements to mitigate this.[11] There was likely variation between study sites in assignment of diagnoses including AARS, however consistency was provided by study investigator and study spine surgeon review of imaging reports and consultant notes for final injury assignment. There was no standardized information about the radiographic criteria for diagnosis or the severity of AARS in these patients and the association with treatment. This subanalysis is from a subset of the main study population, therefore, specific information that may be important to the functional outcome of AARS may be lacking. Nonetheless, this is a large multi-centered cohort of children with AARS with comparison groups, which has allowed us to make interesting observations regarding the presentation, management and outcomes of children with AARS.

Conclusion

AARS should be included in the differential diagnosis of young children with blunt trauma, torticollis and neck pain. Torticollis rarely accompanies other cervical injuries. CT imaging was commonly performed to aid diagnosis. Most children had good outcomes with conservative management. Future studies should aim to identify those children in need of CT imaging and surgical intervention.

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Table 1

Characteristics of the AARS, other cervical spine injuries (CSI) and non-CSI controls.

Characteristic	AARS N = 55	Other CSI N = 485	P-Value*	Non-CSI Controls N = 1060	P-Value*
Age, Mean +/− SD	7.7 +/− 3.8	10.7 +/− 4.6	<0.01	8.9 +/− 4.8	0.06
0 to < 2 y.o.	2 (4%)	25 (5%)		116 (11%)	
2 to < 8 y.o.	27 (49%)	113 (23%)		318 (30%)	
8 to < 16 y.o.	26 (47%)	347 (72%)		626 (59%)	
Male	26 (47%)	318 (66%)	0.01	634 (60%)	0.07
Race			0.51		0.01
White	34 (62%)	298 (61%)		497 (47%)	
Black	7 (13%)	87 (18%)		280 (26%)	
Other	6 (11%)	31 (6%)		51 (5%)	
Not Documented	8 (15%)	69 (14%)		232 (22%)	
Transported by EMS from the scene	26 (47%)	338 (70%)	<0.01	777 (73%)	<0.01
Transfer from outside hospital	36 (65%)	261 (54%)	0.12	205 (19%)	<0.01
Delay in presentation >= 24 hours	11 (21%)	51 (11%)	0.04	14 (1%)	<0.01

[*] Wilcoxon p-value for age; Fisher's exact test p-value for all other comparisons; Not Documented values are not included in calculations of percentages or in statistical tests except for race.

Table 2

Mechanism of injury for AARS, other cervical spine injuries (CSI) and non-CSI controls

Mechanism	**AARS N = 55**	**Other CSI N = 485**	**Non-CSI Controls[‡] N = 1060**
Falls (total) [*]	20 (36%)	90 (19%)	314 (30%)
Elevation [*]	15 (27%)	60 (12%)	202 (19%)
Stairs	1 (2%)	8 (2%)	54 (5%)
Ground level	4 (7%)	22 (5%)	58 (5%)
Motor vehicle crash occupant [*†]	6 (11%)	146 (30%)	260 (25%)
Blunt injury [*]	6 (11%)	10 (2%)	73 (7%)
Sports	5 (9%)	85 (18%)	104 (10%)
Other motorized transport crash (e.g. motorcycle, all terrain vehicle, etc.)	3 (5%)	15 (3%)	32 (3%)
Non-motorized transport collision or fall (e.g. bicycle, scooter, etc.)	2 (4%)	27 (6%)	46 (4%)
Non-motorized vehicle hit by motor vehicle	1 (2%)	15 (3%)	63 (6%)
Pedestrian hit by motor vehicle [*†]	0 (0%)	38 (8%)	123 (12%)
Diving [*]	0 (0%)	35 (7%)	1 (0%)
Hanging	0 (0%)	0 (0%)	2 (0%)
Other [*†]	12 (22%)	24 (5%)	41 (4%)

[*] Fisher's exact test p-value <0.05 comparing AARS to Other CSI

[†] Fisher's exact test p-value <0.05 comparing AARS to Non-CSI controls

[‡] One Non-CSI control patient was missing documentation for injury mechanism and was not included in Fisher's exact tests

Table 3

Clinical characteristics of AARS injuries, other cervical spine injuries (CSI) and non-CSI controls.*

Clinical Characteristic	AARS N = 55	Other CSI N = 485	P-Value	Non-CSI Controls N = 1060	P-Value
Complaint of neck pain	35 (67%)	218 (47%)	0.01	327 (33%)	<0.01
Neck tenderness	28 (53%)	193 (42%)	0.14	369 (36%)	0.02
Torticollis	29 (57%)	21 (5%)	<0.01	55 (6%)	<0.01
Focal neurological findings	7 (14%)	169 (36%)	<0.01	56 (6%)	0.04
Altered mental status	9 (17%)	142 (30%)	0.04	175 (17%)	1.00
Loss of consciousness	12(24%)	189 (43%)	0.01	318 (33%)	0.22
Intubated	4 (7%)	109 (22%)	0.01	74 (7%)	0.79
Substantial head injury	3 (5%)	88 (19%)	0.01	122 (12%)	0.19
Substantial torso injury	3 (5%)	48 (10%)	0.34	56 (5%)	1.00
Substantial extremity injury	2 (4%)	47 (10%)	0.21	93 (9%)	0.22

[*] Not Documented values are not included in calculations of percentages or in statistical tests; all p-values are from Fisher's exact tests of homogeneity.

Table 4

Imaging and interventions performed for AARS and other cervical spine injuries (CSI).

	AARS N = 55	**Other CSI N = 485**	**P-Value**[*]
Plain films	36 (65%)	409 (84%)	<0.01
CT performed	52 (95%)	312 (64%)	<0.01
MRI performed	13 (24%)	284 (59%)	<0.01
Both CT and MRI performed	13 (24%)	180 (37%)	
Neither CT nor MRI performed	3 (5%)	69 (14%)	
Highest level of intervention:			<0.01
None	6 (11%)	80 (16%)	
Soft collar	4 (7%)	29 (6%)	
Rigid collar	20 (36%)	203 (42%)	
Brace	0 (0%)	22 (5%)	
Traction	14 (25%)	0 (0%)	
Halo	9 (16%)	68 (14%)	
Internal fixation	2 (4%)	83 (17%)	

[*] P-values from Fisher's exact test of homogeneity

Table 5

Outcomes for AARS injuries, other cervical spine injuries (CSI) and non-CSI controls.

Outcome	AARS N = 55	Other CSI N = 485	P-Value*	Non-CSI Controls N = 1060	P-Value*
Normal	49 (89%)	337 (69%)	<0.01	1012 (96%)	0.03
Persistent neurological deficit	6 (11%)	108 (22%)		37 (3%)	
Moderate to severe cognitive deficit	4 (7%)	41 (8%)	1.00	22 (2%)	0.04
Dependent ambulation or immobile	3 (5%)	82 (17%)	0.03	22 (2%)	0.12
Incontinent bowel or bladder function OR chronic catheterization	0 (0%)	46 (9%)	0.01	8 (1%)	1.00
Death	0 (0)	40 (8%)		11 (1%)	

[*] P-values from Fisher’s exact test of homogeneity