Editorial

Traumatic atlantoaxial rotatory subluxation in adults: is cervical fusion the answer?

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Introduction

Adult acute atlantoaxial rotatory subluxation (AARS) is an uncommon, yet potentially life-threatening condition characterized by injury to the C1-C2 articulation with disruption of the facet joints and/or surrounding stabilizing ligaments including the alar and transverse ligaments (1). AARS is more commonly seen in younger patients with some studies indicating that the incidence occurs in patients under age 30 and over age 30 at a ratio 2:1, respectively (2). In addition, AARS has been shown to occur three times as frequently in female patients than males (2). Typically, patients sustain these injuries with higher-energy injury mechanisms including motor vehicle collisions and sports-related accidents (3). Although neurologic injury can be catastrophic with cord injury occurring at the level of C1-C2, such neurologic injuries are exceedingly rare and previous studies have shown a low incidence of neurologic deficit in patients with AARS (1,2,4-8). Previous reports have shown only rare instances in which patients have any significant upper spinal cord compression or myelopathy (9-11).

Atlantoaxial rotatory subluxation is a pathologic entity more commonly seen in the pediatric population and resultantly, there is more literature on the topic in this population than there is on adults. Skeletal immaturity in pediatric patients predisposes this population to this injury due to more horizontal geometry of the atlantoaxial joint, a relatively larger head:torso ratio than adults, increased ligamentous laxity and less developed neck musculature. While many of these factors change over time with growth, these features may persist into adulthood for patients. Because this condition is so rare, we currently have only minimal low-level evidence to inform decisions about management. Due to the scarcity of these cases, gathering meaningful evidence from even a retrospective review would likely require a longstanding multi-center study. Spirollari *et al.* (12) utilized a retrospective study to better understand the prevalence of neurologic deficit and report on patient outcomes with traumatic AARS. Drawing from the National Inpatient Sample database, this study was elicited useful data regarding outcomes and complications of operative and nonoperative management for these patients.

Study overview

This study was a retrospective cohort study of patients from the National Inpatient Sample Database from 2016–2019 (12). ICD-10 diagnosis codes for atlantoaxial rotatory subluxation were used to identify patients who were then sorted into operative and nonoperative management cohorts. Of 990 adult patients with AARS were identified in total and this population was split into two cohorts of AARS patients; 270 (27.3%) undergoing operative management with cervical fusion (CF) and 720 (72.7%) undergoing nonoperative management. The patients undergoing CF had a higher rate of myelopathy and lower inpatient mortality rates. CF patients were found to have a longer length of stay and increased healthcare resource utilization. Patients managed nonoperatively had more severe injury scores and CF patients had lower injury severity.

Strengths & weaknesses

The most important strength is also the weakness of this study. Because AARS is such a rare condition with only minimal low-level evidence available to guide decision-making, this study has inherent value because it does attempt to address a void in the literature by leveraging the high volume of cases afforded by a national database. This is a dual-edged sword as database studies, particularly of more rare and unusual conditions, can be subject to significant bias due to the methodology of how it is documented. In addition, with a retrospective study design, there may be clear selection bias in which patients should undergo surgical fixation. Although in an ideal world one could design a prospective randomized controlled trial comparing the two cohorts of interest using standardized protocols surrounding surgical and nonoperative treatment, this simply is not possible currently with the low volume of these cases.

Discussion

At face value, this study indicates that patients with AARS managed surgically with posterior CF have better surgical outcomes overall. However, as the authors point out, this finding likely is more of a reflection of selection bias that drives surgeons towards one treatment option or another. It is well-documented that surgeons tend to withhold more aggressive surgical treatments for patients with more severe injuries or poorer prognoses. Therefore, it is unsurprising that the patients in the nonoperative cohort were found to have a higher injury severity score and poorer outcome scores than patients in the operative cohort with a statistically significant difference. This selection bias also highlights the importance of improving preoperative risk stratification beyond "preop clearance", but to incorporate some element of prognostication for the polytraumatized patient with spinal injuries. It is possible that the decision-making which leads certain patients to fall into the "nonoperative" category is based on inappropriate or inaccurate prognostication. This is an area where more research is warranted.

As healthcare enters the era of 'value-based care', providers' medical decision-making surrounding the management of conditions such as AARS will come under increased scrutiny if there is a lack of strong evidence to support one treatment over another. Although it may not be possible to draw decisive conclusions by comparing the two cohorts head-to-head, looking at the groups separately does provide useful data to add to our knowledge of this condition—the study is a large cohort of patients bearing a diagnosis of AARS and there is an exceedingly low rate of neurologic injury, there is a low rate of surgical complications (lower than historical reports of C1-2 posterior CF, potentially reflecting improved technology/techniques).

The authors should be commended on performing a study on a rare condition in which surgical management is not fully understood.

Conclusions

Spirollari *et al.* (12) demonstrated an incidence of AARS in 990 patients over a three-year period, with those undergoing CF have higher hospital costs with decreased mortality. This study, while limited due to its methodology, offers pertinent information about AARS and how treatment must be considered in the setting of the polytrauma patient.

The most notable finding of this study was surrounding the injury severity of patients in the two cohorts. The more severely injured patients were more likely to be managed nonoperatively and this reflects a known bias amongst surgeons to withhold more aggressive and invasive treatment for patients with more severe injuries. This is an important issue to explore as injury severity does not necessarily correlate with or predict a patient's prognosis or future candidacy for a potential surgical intervention. In the setting of trauma, it is not uncommon for patients to present with severe injuries including traumatic brain injuries and low Glasgow Coma Scales yet still have remarkable recovery to near baseline function. During the early care of polytraumatized patients, it is often very hard to anticipate the recovery, therefore longer follow-up of these non-operative patients may help decide how many are successfully managed without fusion. Patients managed surgically were found to have a lower mortality however this cohort had a lower injury severity than the nonoperative cohort—again reflecting the impact of the selection bias.

Next steps

Future national database studies on this topic can build upon the work done by this group by further refining the research question and patient selection as diagnosis and procedure coding for patients becomes more detailed, sophisticated, and accurate. This is particularly important in spine surgery research as the approach to diagnosis and treatment is very detailed and patient-specific; much of this detail is simply not captured by the major national databases currently

available. This paper uses the ICD-10 diagnosis code for AARS and the Current Procedural Terminology (CPT) codes for various generic posterior spinal decompression and fusion techniques. Most likely, these patients are having the surgeries represented by these CPT codes to treat AARS but there is no way to know the true indication for surgery in these patients. It is possible that some of them had posterior cervical decompression and fusion for the indication of cervical myelopathy and they carry a diagnosis of AARS but it was not the true indication for surgery.

Because of the significant selection bias at play in comparison of these two cohorts, more research to inform management is needed. Since it is not realistic to expect a prospective randomized control trial to be done on this topic, it will be important to find creative ways to help minimize the bias that occurs with these types of studies. Again, the authors should be commended on adding valuable information on a rare, potentially life-threatening injury that spine surgeons must be prepared to face.

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Footnote

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Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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