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Atraumatic atlantoaxial subluxation in pediatric enthesitis-related juvenile idiopathic arthritis: illustrative case

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BACKGROUND Juvenile idiopathic arthritis (JIA) is the most common pediatric rheumatological disease, yet cervical spine involvement remains an underrecognized but potentially devastating manifestation. Atlantoaxial subluxation (AAS) arises from inflammatory changes causing ligamentous laxity and instability.

OBSERVATIONS A 13-year-old female presented with progressive neck pain. Imaging revealed a 10-mm atlantodental interval on CT, along with hyperintensity and stretching of the transverse atlantal ligament on MRI. She underwent a posterior C1–2 open reduction and fusion. Subsequent rheumatological workup confirmed enthesitis-related JIA, based on polyarticular arthritis, HLA-B27 positivity, and elevated inflammatory markers. To contextualize this case, the authors performed a systematic review and meta-analysis of JIA-related AAS across 21 studies. The pooled incidence of AAS was 14%, with a mean age at JIA onset of 8.47 years and a female predominance of 62%. Enthesitis-related arthritis emerged as the most frequently reported subtype, and 94.4% of patients with AAS improved posttreatment.

LESSONS This case and supporting literature underscore the importance of early detection and multidisciplinary management of AAS in pediatric patients with JIA. Timely neurosurgical stabilization, combined with optimized immunosuppressive therapy, can prevent neurological compromise. Future research should focus on standardized diagnostic thresholds and outcome measures to guide best practices.

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KEYWORDS atlantoaxial subluxation; juvenile idiopathic arthritis; enthesitis-related arthritis; pediatric spine instability; cervical spine fusion

While adult rheumatoid arthritis (RA) has been extensively associated with cervical spine pathology, few studies have investigated this association in the pediatric population.[1–6] RA is among the most widespread chronic illnesses in children, with an annual incidence of 1.6–23 per 100,000 adolescents and an annual incidence of approximately 1 to every 1000 developing the juvenile idiopathic arthritis (JIA) type of chronic arthritis.[7] JIA unifies all manifestations of chronic childhood arthritis and has been split into subtypes based on the number of affected joints and differing genetic susceptibility: oligoarticular (persistent or extended, ≤ 4 joints), polyarticular (rheumatoid factor [RF] negative or RF positive, ≥ 5 joints affected), systemic, psoriatic, enthesitis-related, or undifferentiated (does not fit into one of the categories or corresponds to > 1 subtype).[8]

Enthesitis-related arthritis (ERA) is often associated with HLA-B27 and uveitis, and it involves heightened expression of Toll-like receptors 2 and 4 on monocytes, leading to proinflammatory cytokine production.[8–11] Elevated cytokines in ERA include monocyte-derived interleukin (IL)–1 and IL-6 in synovial fluid and an increase in Th17 cells, which drive inflammation via the IL-23/IL-17 axis.[8,12] Mechanical stress on stromal cells at entheseal sites and direct IL-23 action initiate enthesitis, which can culminate in RANK-expressing osteoclast-mediated bone erosion, soft tissue calcifications, and new bone formation.[8,12] Spinal facet joints, also synovial, are similarly affected by this pathogenic process. The atlantoaxial complex is uniquely prone to instability through immune-mediated damage due to its 3 synovial joints lacking an intervertebral disc, its heavy reliance on dense ligamentous support (rich in type I and III collagen), and its high degree of mobility.[13] Structural anomalies, such as occipitalization of the atlas, may further amplify biomechanical stress at this junction, accelerating destabilization.[14]

ABBREVIATIONS AAS = atlantoaxial subluxation; ADI = atlantodental interval; ANA = antinuclear antibody; ERA = enthesitis-related arthritis; ESR = erythrocyte sedimentation rate; IL = interleukin; JIA = juvenile idiopathic arthritis; NSAID = nonsteroidal anti-inflammatory drug; RA = rheumatoid arthritis; RF = rheumatoid factor.

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ERA-induced cervical spine instability can impinge on the cervical spinal cord, and cause paralysis.[15,16] The further the atlantodental (ADI) interval, the increased severity of the pathology and the increased likelihood of mortality.[16] Given these potential catastrophic outcomes, prompt neurosurgical evaluation and possible stabilization are essential for preventing irreversible neurological damage in pediatric patients in whom conservative measures have failed or those who exhibit significant instability. There are a limited number of cases of ERA associated with cervical spine atlantoaxial subluxation (AAS) in adolescents in the literature. Here, we present a case of atraumatic AAS in a pediatric patient with JIA. We also review lessons learned from the published literature on AAS and JIA.

Illustrative Case

A 13-year-old female with no history of trauma presented to the emergency department (ED) after 3 months of progressively worsening intermittent neck pain. She initially saw a chiropractor and underwent neck adjustments without significant change in symptoms. On the day of admission, she described a severe exacerbation of pain (10/10) in the posterior cervical region, radiating to both shoulders, along with intermittent shoulder paresthesias and transient right thumb numbness. She also reported 1 year of intermittent pain and swelling in her left middle finger.

Physical examination revealed torticollis and tenderness over the bilateral posterolateral cervical musculature, but normal strength and intact sensation in all extremities. CT of the cervical spine (Fig. 1A and B) demonstrated abnormal widening of the C1–2 interval with anterior subluxation of the atlas on the axis measuring 10 mm and narrowing of the spinal canal with posterior atlantodental index of 10 mm. MRI (Fig. 1C and D) confirmed posterior displacement of the dens relative to C1, nearly complete effacement of the CSF at C1–2, and soft tissue enhancement and T2 hyperintensity. Laboratory studies were remarkable for an erythrocyte sedimentation rate (ESR) of 36 mm/hr and leukopenia of 3.33/μL.

The patient's history, physical examinations, and imaging findings were suggestive of an inflammatory process, raising suspicion for JIA. Given the progressive atlantoaxial instability, ongoing pain, and concern for cord compression in the context of inflammatory pannus and moderate canal stenosis, surgical intervention was favored over observation or prolonged medical management. Although conservative therapy with a cervical collar provided temporary symptom relief, the radiographic evidence of 1.1-cm subluxation, pannus formation, and near-complete effacement of the ventral and dorsal CSF spaces suggested a high risk of catastrophic neurological deterioration.

One month after initial presentation, the patient underwent C1–2 posterior decompression and fusion. Operative risks, including bleeding, infection, vertebral artery injury, potential for nerve root sacrifice with possible dysesthesia, hardware failure, and spinal cord injury, were discussed extensively with the family. Preoperative CT angiography of the head and neck demonstrated normal vertebral artery anatomy and no hemodynamically significant stenosis, supporting safe planning for instrumentation. The patient was secured in Mayfield pins for cranial fixation, placed supine on an OSI table (Mizuho OSI) with the neck in neutral/slight lordosis, and turned prone. A lateral radiograph obtained after positioning showed 30% reduction of ADI. During this initial reduction, the mean arterial pressure dropped to 50 mm Hg, associated with decreased motor evoked potentials more so in the bilateral upper extremities than in the bilateral lower extremities. These resolved after hemodynamic augmentation and steroid administration. Bilateral C2 nerve roots were sacrificed to optimize exposure and arthrodesis. A C1 laminectomy was performed to decompress the spinal canal, facilitate reduction, and allow room for graft placement while avoiding cord compression in a canal already narrowed by inflammatory pannus. A C1 lateral mass screw and C2 pedicle screws were placed under intraoperative navigation. Intraoperative CT showed complete reduction of the AAS with ADI of 3 mm. There were no complications, and the patient was discharged on postoperative day 2. She reported improved neck pain and bilateral upper extremity paresthesias.

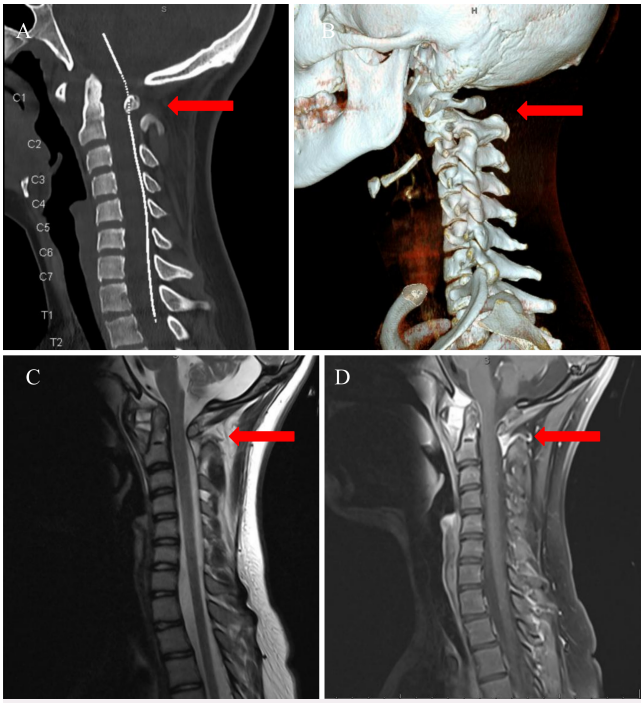

FIG. 1. Abnormal widening of the C1–2 joint space with anterior displacement of C1 relative to C2, measuring 10 mm (*red arrows* in panels A and B), resulting in mild narrowing of the thecal sac to 8 mm anteroposteriorly in the supine position. **A:** Sagittal cervical spine soft tissue image. **B:** Sagittal 3D anatomical reconstruction. No associated fracture is observed. **C and D:** T2-weighted (C) and T1-weighted (D) cervical spine images revealing abnormal widening of the anterior C1–2 interval, along with posterior displacement of the dens relative to C1 and the skull base. This led to mild thecal sac narrowing and near-complete effacement of the CSF (*red arrows*). The abnormal soft tissue within the C1–2 space exhibits postcontrast enhancement and T2 hyperintensity, suggestive of inflammatory soft tissue, with a differential diagnosis including pannus related to JIA.

Over the subsequent 5 months, the patient began exhibiting progressive joint involvement of the temporomandibular joints, elbows, wrists, metacarpophalangeal joints, proximal interphalangeal joints, knees, and ankles, coupled with enthesitis. Rheumatological evaluation revealed HLA-B27 positive, ANA positive, rheumatoid factor negative, confirming diagnosis of the enthesitis-related subtype of JIA. She was started on an intensified immunosuppressive regimen, including methotrexate, adalimumab (Humira), and a prednisone taper. At her most recent follow-up, approximately 6 months after her initial presentation, she reported significant relief of pain, improved function, and a notable decrease in inflammatory markers. Repeat cervical imaging revealed stable hardware alignment without recurrence of subluxation

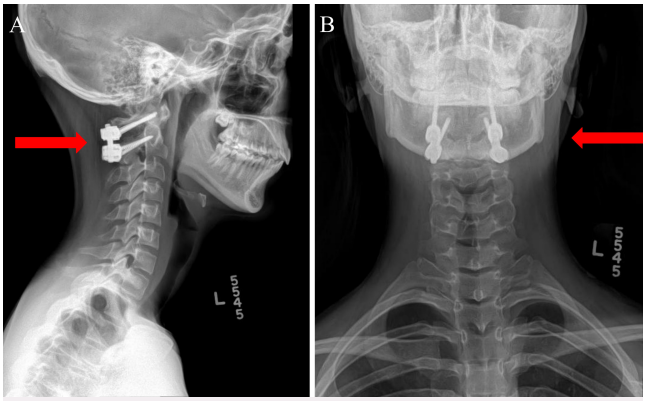

FIG. 2. Six-month follow-up lateral (**A**) and anteroposterior (**B**) radiographs of the cervical spine, showing C1 laminectomy and posterior fusion of C1–2 with stable hardware alignment (*red arrows*). No paravertebral lucency is observed, cervical lordosis remains straightened, and the previously noted C1–2 widening has been corrected.

(Fig. 2). Although she continued to experience mild headaches and numbness in the C2 dermatome, she retained approximately 50%–60% of normal cervical range of motion, sufficient for daily activities.

Systematic Review

A systematic review was conducted using PubMed, ScienceDirect, and Scopus, following PRISMA guidelines. The following MeSH search terms "juvenile idiopathic arthritis" AND "atlantoaxial subluxation" were combined using Boolean operators, with no timeframe limit. We included original case reports presenting primary information such as risk factors, clinical presentation, management, and outcomes of JIA associated with AAS. Literature reviews, correspondence, commentaries, letters to the editor, book chapters, animal studies, opinion pieces, and systematic reviews and meta-analyses were excluded.

Three authors (S.A., S.O., and T.Y.P.) independently conducted the initial screening process based on titles and abstracts to determine eligibility. These authors then reviewed the full texts to finalize the inclusion and extract relevant data. The bibliographies of included articles were also examined for additional relevant studies. Disagreements were resolved through consensus meetings. The selected articles were assessed using our data extraction methodology. Key variables included 1) demographics, 2) medical history and clinical presentation, 3) diagnostic modalities, 4) JIA subtypes, 5) medications, 6) conservative and surgical management strategies, and 7) outcome. Outcomes reported were clinical improvement, persistent symptoms, or other relevant clinical endpoints at the last follow-up. We also documented study limitations, conclusions, and bibliometric details such as study design, publication year, and country.

Descriptive statistics were used to categorize demographics, presentation, management, and outcome measures across the included cases. Mean values and standard deviations were calculated for continuous variables, while categorical variables were summarized using frequencies and percentages. For retrospective cohort studies, a meta-analysis was performed using random-effects models to account for between-study variability. The incidence of AAS was pooled using the metaprop function from the meta-R package. For studies reporting age at JIA onset, the pooled mean age was estimated using the metamean function.

Our systematic literature review identified 623 citations through database searches and 45 through other peer-reviewed sources (Fig. 3). Fifty-two citations were removed due to being duplicates; 616 citations were then screened in full for eligibility, and 595 were excluded. Ultimately, 21 studies met inclusion criteria, consisting of 15 case reports with 19 patients (Table 1)[17–31] and 6 retrospective studies with 466 patients (Table 2).[32–37]

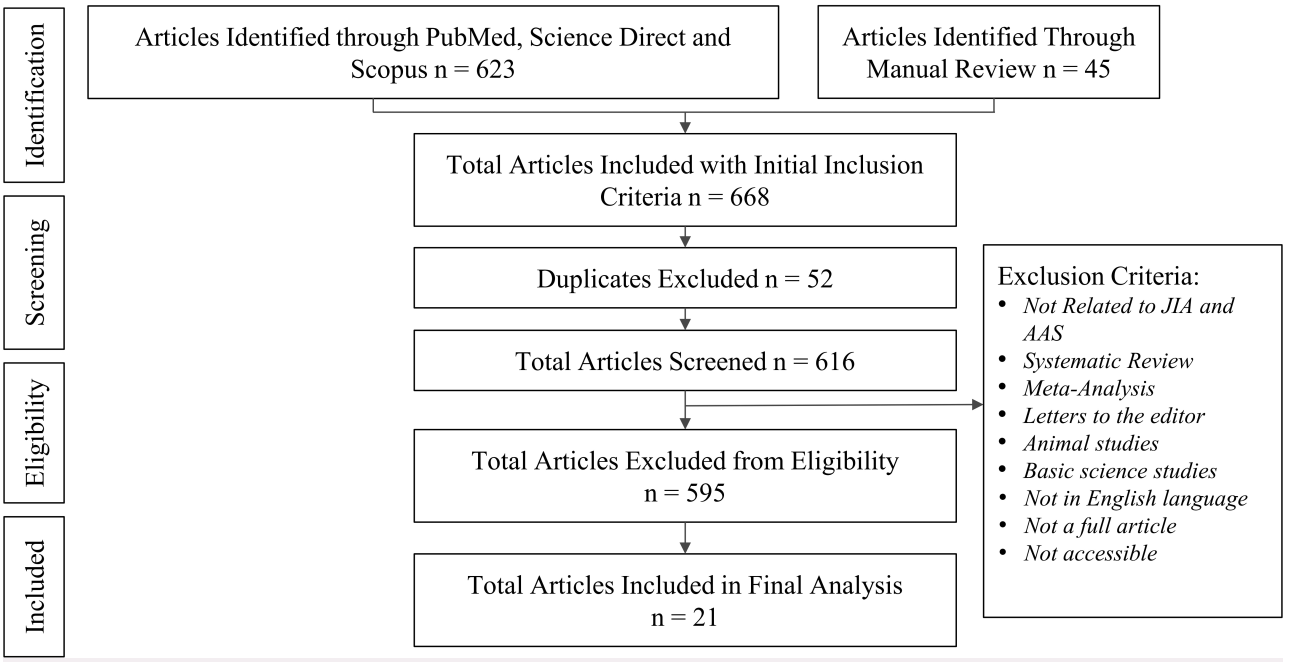

FIG. 3. PRISMA flow diagram showing the methodology employed in conducting the systemic review.

Of the 19 patients with JIA and AAS identified in 15 case reports, the mean age at presentation was 10.8 ± 3.7 years, and most had no history of trauma (78.9%, n = 15/19). A male predominance (12 males, 7 females) was observed in this case report cohort. Only 21.1% (n = 4/19) had an inciting traumatic event preceding AAS development.

Notably, the enthesitis-related subtype was the most frequently reported (86.7%, n = 13/15) when subtypes were specified, with most patients having HLA-B27 positivity with elevated inflammatory markers (mean ESR 44.8 ± 38.3 mm/hr). The mean ADI was 8.2 ± 2.9 mm, indicating significant C1–2 instability.

Therapeutic strategies for these 19 patients encompassed both surgical and conservative interventions. In total, 57.9% (n = 11/19) underwent C1–2 posterior fusion; within this subgroup, 54.5% (n = 6/11) also used cervical collars, 63.6% (n = 7/11) underwent traction, and 18.2% (n = 2/11) used a Halo vest. The remaining 42.1% (n = 8/19) were managed conservatively; of these, 62.5% (n = 5/8) used cervical collars and 12.5% (n = 1/8) underwent some form of traction. Medications varied, including nonsteroidal anti-inflammatory drugs (NSAIDs), steroids, disease-modifying antirheumatic drugs, biologics, and adjuncts such as acetaminophen, diazepam, and folic acid. Overall, 94.4% (n = 17/18) of the reported cases demonstrated clinical improvement, whereas 5.6% (n = 1/18) progressed to persistent ankylosis. Our patient, a female with HLA-B27 negativity who was managed only conservatively, initially presented with spontaneous AAS and progressed to ankylosis despite conservative management with a cervical collar, NSAIDs, methotrexate, sulfasalazine, and etanercept.[25] Despite variability in management, generally progressive instability and neurological comprise prompts surgical intervention, while milder cases are often managed conservatively.

A random-effects meta-analysis of 6 retrospective cohort studies (totaling 473 patients with JIA) estimated the pooled mean age of JIA onset at 8.47 years (95% CI 6.88–10.07 years, 3 studies), with a female predominance (62%, 95% CI 30%–86%, 4 studies), a divergence from the largely male skew in case reports. Critically, the pooled incidence of AAS in these studies was 14% (95% CI 6%–28%, 6 studies), suggesting that approximately 1 in 7 JIA patients might develop this serious cervical spine instability. The I^2 heterogeneity values for age, sex, and incidence analyses (86.5%, 95%, and 86.3%, respectively) underscore significant variability in demographic profiles and diagnostic thresholds across the literature.

Limitations

Despite providing valuable insights, this study has several limitations that must be acknowledged. The relatively small number of published case reports and retrospective studies restricts the generalizability of our findings and highlights the need for larger multicenter or prospective investigations to validate observed trends. Furthermore, relying heavily on small case series and case reports introduces a risk of publication bias toward favorable outcomes; this phenomenon, previously noted in neurosurgical research, can skew complication rates and underrepresent adverse events.[38] Variations in diagnostic criteria, imaging protocols, and follow-up intervals across studies further complicate direct comparisons. The retrospective nature of included data also raises concerns about incomplete documentation and recall bias. Additionally, because many of these retrospective studies focused primarily on JIA patient characteristics rather than the specific clinical trajectories of those with AAS, our capacity for in-depth outcome driven meta-analysis in this subset was constrained. Finally, heterogeneity in treatment regimens, including various medication protocols, limits the ability to draw definitive conclusions regarding optimal management strategies for pediatric AAS in JIA.

Informed Consent

The necessary informed consent was obtained in this study.

Discussion

Observations

AAS is a potentially severe complication of JIA, particularly in patients with the ERA subtype. Our meta-analysis indicates that cervical spine pathology appears to be more common in JIA than previously appreciated, with retrospective data highlighting its significant prevalence. For instance, Kotecki et al. found that 35% of children with JIA developed at least one radiographic lesion in the cervical spine, emphasizing the need for vigilant monitoring.[32] Among the most severe manifestations is AAS, which requires careful management to prevent progressive instability. While surgical fusion has been performed in select pediatric cases, its long-term consequences, such as reduced cervical mobility and potential adjacent-level stresses, must be balanced against the risks of untreated instability. Beier et al. reported a 12.5% fusion rate among JIA patients, underscoring the necessity for larger cohort studies to refine surgical indications and improve management strategies for this vulnerable population.[39]

Conservative management, such as cervical collar immobilization, physical therapy, and anti-inflammatory medications, is typically the initial approach for cervical spine involvement. However, our systematic review and meta-analysis suggest that while many patients respond favorably to these measures, a significant subset, especially those with severe instability or neurological involvement, require neurosurgical intervention. Notably, more than half of the case report patients analyzed in our review underwent a C1–2 posterior fusion procedure and 94.4% of all reported cases showed clinical improvement postintervention, highlighting the importance of early recognition and timely escalation of care. C1–2 arthrodesis demonstrates positive outcomes in pediatric patients, including high fusion rates, improved neurological status, and minimal complications.[40] Posterior atlantoaxial screw fixation, including partial removal of the C1 lamina, if necessary, can effectively relieve cord compression and achieve stable fusion in at-risk populations such as children with Down syndrome.[41]

Our systematic review and meta-analysis reveal a higher-than-anticipated incidence (14%) of AAS in pediatric JIA, particularly within the ERA subtype. These findings underscore the importance of early screening for cervical spine involvement, given the high rate of clinical improvement (94.4%) when appropriate interventions are administered. Despite the apparent success of surgical stabilization and immunosuppressive therapy, the included studies exhibit considerable heterogeneity in diagnostic criteria, imaging methods, and treatment thresholds. This variability highlights the urgent need for standardized approaches to optimize patient outcomes and guide future research on the prevention and management of AAS in pediatric JIA.

In select cases, dynamic imaging such as flexion-extension radiographs or dynamic MRI can offer additional diagnostic clarity, particularly when instability is equivocal on static imaging.[42] However, in our patient, cervical spine CT and MRI revealed clear evidence of atlantoaxial instability, including a 1.1-cm ADI, pannus formation, and near-complete effacement of the CSF space. Given the inflammatory etiology, risk of cord compression, and instability already evident on static imaging, additional dynamic studies were deemed unnecessary and potentially hazardous. Thus, the diagnosis and surgical planning

TABLE 1. Patient characteristics, management strategies, and outcomes at last follow-up for all case reports included in the systematic review

Authors & Year	Age (yrs), Sex	Comorbidities	Hx of Trauma	HLA-B27	ANA	RF	CRP, mg/dL	ESR, mm/hr	JIA Subtype	MRI	CT	ADI, mm	Medications	C1–2 Pst Fusion, CC/HT	Outcome
Siu et al., 2023[17]	16, M	Asthma	No	Yes	No	No	0.46	11	Enthesitis related	Yes	Yes	NS	Naproxen, sulfasalazine, acetaminophen	No, CC	Improved
	8, M	None	Yes	Yes	No	No	3.09	116	Enthesitis related	Yes	Yes	4.4	Naproxen, methylprednisolone, prednisolone, methotrexate, sulfasalazine, folic acid	No, CC & halter traction	Improved
Whitaker & Glotzbecker, 2021[18]	5, F	None	No	No	No	No	0.1	8	NS	Yes	Yes	NS	Sulfasalazine, etanercept, pulse steroids, diazepam	Yes, CC & HT	Improved
	8, F	None	No	Yes	Yes	No	0.4	13	NS	Yes	Yes	NS	Methotrexate, steroids, mycophenolate mofetil, infliximab	Yes, CC & HT	Improved
	9, F	None	No	No	Yes	No	0.1	13	NS	Yes	Yes	NS	Infliximab, methylprednisolone	Yes, CC, HT, & halo vest	Improved
La Tessa et al., 2020[19]	13, M	None	No	Yes	No	No	NS	NS	Enthesitis related	Yes	Yes	NS	Methylprednisolone	Yes, none	Improved
Prada et al., 2018[20]	10, F	Down syndrome	No	Yes			0.1	13	Enthesitis related	Yes	Yes	11	Methylprednisolone, infliximab, oral anti-inflammatory medications	Yes, HT & halo vest	Improved
Enazi et al., 2014[21]	12, F	None	Yes	Yes	No	No	NS	NS	Enthesitis related	Yes	No	5	Naproxen, prednisone, methotrexate, sulfasalazine, etanercept	Yes, none	Improved
Salem et al., 2014[22]	10, M	Cerebral palsy	Yes	NS	NS	NS	NS	NS	NS	Yes	Yes	2.3–11.4	Methylprednisolone, methotrexate	Yes, HT	Improved
Taddio et al., 2011[23]	8, F	None	No	No	No	No	NS	NS	Oligoarticular	Yes	Yes	NS	Methylprednisolone, methotrexate, infliximab	No, none	Improved
Kobayashi et al., 2006[24]	13, M	None	No	Yes	No	No	1.5	64	Enthesitis related	Yes	No	11	NSAID, dexamethasone, methotrexate, sulfasalazine	No, none	Improved
Breda et al., 2005[25]	7, F	None	No	No	No	No	NS	30–40	Enthesitis related	No	No	NS	Naproxen, methotrexate, sulfasalazine, etanercept	No, CC	Persistent ankylosis
Mitra et al., 1999[26]	6, M	None	No	No	NS	NS	NS	Elevated	Systemic onset	No	Yes	2–8	Naproxen	No, CC	Improved
Haasbeek & Lessard, 1998[27]	9, M	None	No	Yes	No	No	NS	Elevated	Enthesitis related	No	Yes	>10	Aspirin, naproxen, ocular steroids	Yes, CC & halter traction	Improved
Foster et al., 1995[28]	12, M	None	No	Yes	No	No	NS	75	Enthesitis related	Yes	Yes	4–12	Diclofenac	Yes, CC & HT	Improved
	13, M	None	No	Yes	No	No	NS	NS	Enthesitis related	Yes	No	3–8	Tolmetin sodium, sulfasalazine, prednisone	Yes, none	Improved
Kernodle et al., 1987[29]	20, M	None	No	No	No	No	NS	80	Enthesitis related	No	No	7	NS	No, none	NS
Reid & Hill, 1978[30]	15, M	None	No	Yes	No	No	NS	NS	Enthesitis related	No	No	11	NS	No, CC	Improved
Thompson et al., 1982[31]	11, M	None	Yes	Yes	No	No	NS	55	Enthesitis related	No	No	8	Indomethacin	Yes, CC	Improved

CC = cervical collar; CRP = C-reactive protein; HT = halo traction; Hx = history; NS = not specified; pst = posterior.

TABLE 2. Patient characteristics for all retrospective studies included in the systematic review

Authors & Year	No. of Pts	Age, yrs, Mean ± SD (range)	No. of Males (%)	No. of Females (%)	No. w/ AAS/ No. w/ JIA (%)
Fried et al., 1983[34]	92	NA	NA	NA	5/92 (5.4)
Laiho et al., 2002[35]	159	8 ± NA (0.5–15.9)	31 (19.5)	128 (80.5)	27/159 (17)
Laiho et al., 2001[33]	49	NA	NA	NA	17/49 (34.7)
Kjellberg et al., 2011[36]	82	7.4 ± NA (1–16)	21 (25.6)	61 (74.4)	4/82 (4.9)
Elhai et al., 2013[37]	57	10.2 ± 4.6 (NA–NA)	47 (82.5)	10 (17.5)	19/57 (33.3)
Kotecki et al., 2021[32]	34	NA	9 (26.5)	25 (73.5)	2/34 (5.9)

NA = not available; pt = patient.

were based on cross-sectional imaging alone, consistent with practice in cases of known inflammatory subluxation where provocation could exacerbate symptoms or neurological compromise.

Unlike many previously reported cases, our patient experienced a pronounced hemodynamic fluctuation (her mean arterial pressure dropped to 50 mm Hg) and transient reductions in motor evoked potentials in her upper extremities during the initial reduction. These abnormalities resolved rapidly following targeted hemodynamic augmentation and steroid administration, highlighting the importance of neuromonitoring and immediate blood pressure management during high-risk manipulations. Additionally, bilateral C2 nerve roots were sacrificed to optimize exposure for hardware placement and arthrodesis, an approach reserved for complex cases of severe instability. Finally, the coexistence of HLA-B27 and ANA positivity further underscores the heterogeneous immunological profile in JIA-related subluxations.

Lessons

This case underscores the critical need to maintain a high index of suspicion for cervical spine involvement in patients with JIA, especially those presenting with persistent neck pain or neurological symptoms. Prompt recognition and diagnosis are essential to prevent severe complications and guide appropriate management. Future retrospective and prospective studies should focus on identifying clinical and demographic characteristics of JIA patients at heightened risk for AAS, as well as evaluate treatment strategies and their impact on clinical outcomes.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Amasa, Pratt. Acquisition of data: O'Leary, Amasa, Pratt. Analysis and interpretation of data: O'Leary, Patel, Morden, Pratt. Drafting the article: O'Leary, Amasa, Patel. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: O'Leary. Statistical analysis: O'Leary. Administrative/technical/material support: Morden, Pratt. Study supervision: O'Leary, Pratt.

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