A Rare Case of Chronic, Atraumatic Atlantoaxial Rotatory Fixation in an Adult Treated with Halo Traction Suspension, Open Reduction and C1-C2 Fusion

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Introduction

Atlantoaxial rotatory fixation (AARF) is a rare condition in the adult population, with the vast majority being caused by trauma. Non-traumatic cases of AARF are even more rare, with only seven prior documented cases.[1–7]

Given the uncommon nature of this condition in adults, there is no consensus optimal treatment protocol. When diagnosed early, often AARF is treated successfully with non-operative measures such as traction with closed reduction. The diagnosis can be elusive as patients frequently present with normal neurologic exams and non-diagnostic cervical spine radiographs. In those with a delayed diagnosis of AARF, conservative treatment is less successful and patients generally require open reduction and C1-C2 fusion.

This is a case of an adult patient with delayed presentation of atraumatic atlantoaxial rotatory fixation. After informed consent and discussion with his surgeon, he was treated using halo traction suspension with open reduction and posterior C1-C2 fusion.

Case

Presentation

A 61-year-old male software developer with a past medical history significant for controlled type 2 diabetes mellitus, hypertension, nasopharyngeal cysts, and a history of deep vein thrombosis/pulmonary embolism previously treated with anticoagulation therapy presented to the outpatient spine clinic with a chief complaint of progressive torticollis over several years.

The patient was initially seen by physical medicine & rehabilitation in 2016 for left-sided neck tightness and paresthesias in the left ring and small fingers. He underwent a course of physical therapy and a series of cervical epidural steroid injections, with transient resolution of his symptoms. Despite completing multiple non-operative modalities, he continued to have left-sided neck pain and developed a noticeable head tilt. He was referred to see a spine surgeon, but was not seen in a surgical clinic until 2021 when he presented to establish care. At this point his torticollis had worsened. In addition to his complaint of atraumatic chronic neck pain, he endorsed mild tingling in bilateral small fingers without motor weakness. He denied changes in balance, or alterations in fine motor skills.

During examination, a tight left sternocleidomastoid was noted which caused the head to rotate to the right and tilt to the left (Figure 1). He had full strength and intact sensation in bilateral upper and lower extremities.

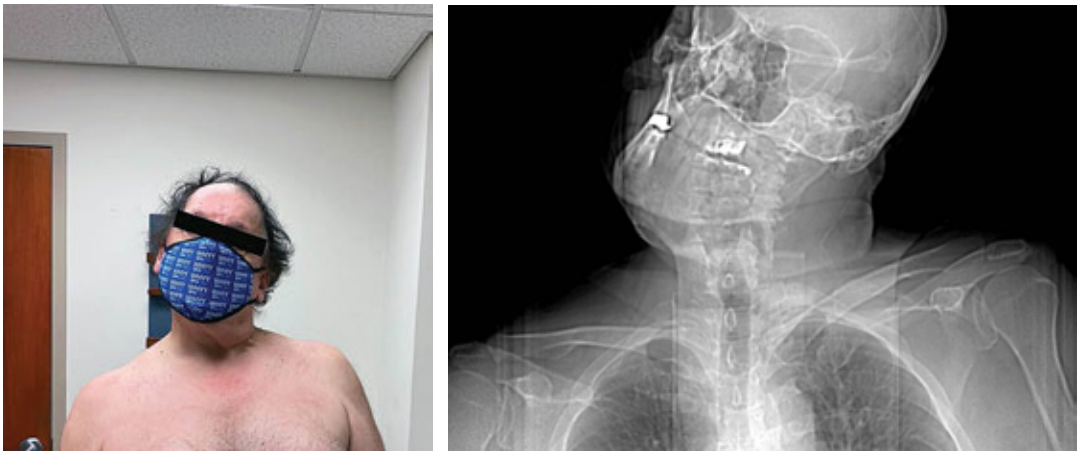

Figure 1. Pre-operative clinical photo and scout CT image demonstrating the classic "Cock-Robin" posture, with the head yawed to the right and rolled to the left.

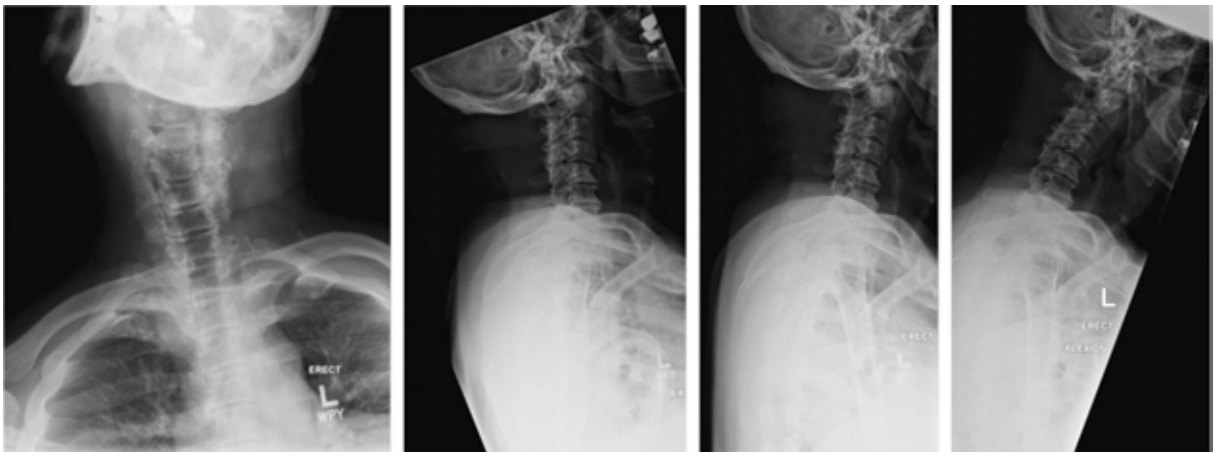

Figure 2. AP and lateral radiographs demonstrating head roll to the left associated with multi-level left-sided spondylosis. No instability is observed on flexion/extension views.

Antero-posterior (AP) and lateral radiographs of the cervical spine showed a head tilt to the left associated with multi-level left-sided spondylosis, without any evidence of instability (Figure 2). a CT scan obtained during evaluation at an outside institution the year prior demonstrated rotatory subluxation of the C1-C2 articulation without anterior displacement, consistent with Fielding Type I rotatory fixation (Figure 3).[8] An open-mouth odontoid view was unavailable for interpretation.

Given the patient presented with a five-year history of this issue and had failed physical therapy and injections, the senior surgeon felt that he would benefit from operative intervention. An MRI was obtained to determine the extent of the operative intervention given his ongoing bilateral hand symptoms. Additionally, a CT angiogram (CTA) was obtained to evaluate the vascular anatomy to plan safe placement of instrumentation.

The MRI showed multilevel cervical stenosis without severe cord compression and the CTA showed intact vertebral arteries without aberrancy. The patient was consented and scheduled for navigation-guided C1-C2 posterior spinal fusion with iliac crest bone graft.

Surgical Management

Following pre-operative optimization for glucose control, the patient was brought to the operating room. A halo was applied under general anesthesia while supine on a stretcher. He was then re-positioned prone on an open Jackson table. The halo was suspended and traction applied, using ten pounds for suspension and ten pounds for traction (Figure 4). A navigation array was then attached to the halo (Figure 5).

Through a posterior midline incision, dissection was carried out to the lateral masses of C1 and C2, taking great care to avoid the vertebral artery laterally in the setting of the patient's distorted anatomy. With fluoroscopy, the C1-C2 joint was localized. Bilateral C2 neurectomies were performed to improve access to the C1-C2 joint, allowing for better release and easier screw insertion later in the case.

To free up the C1-C2 segment, the facet joints were released using curettes and a small sharp Cobb elevator. Once the release was adequate, the halo device and head were manipulated to reduce the rotatory subluxation. With the traction suspension still applied, the reduction maneuver was performed by rotating the head to the left and tilting the

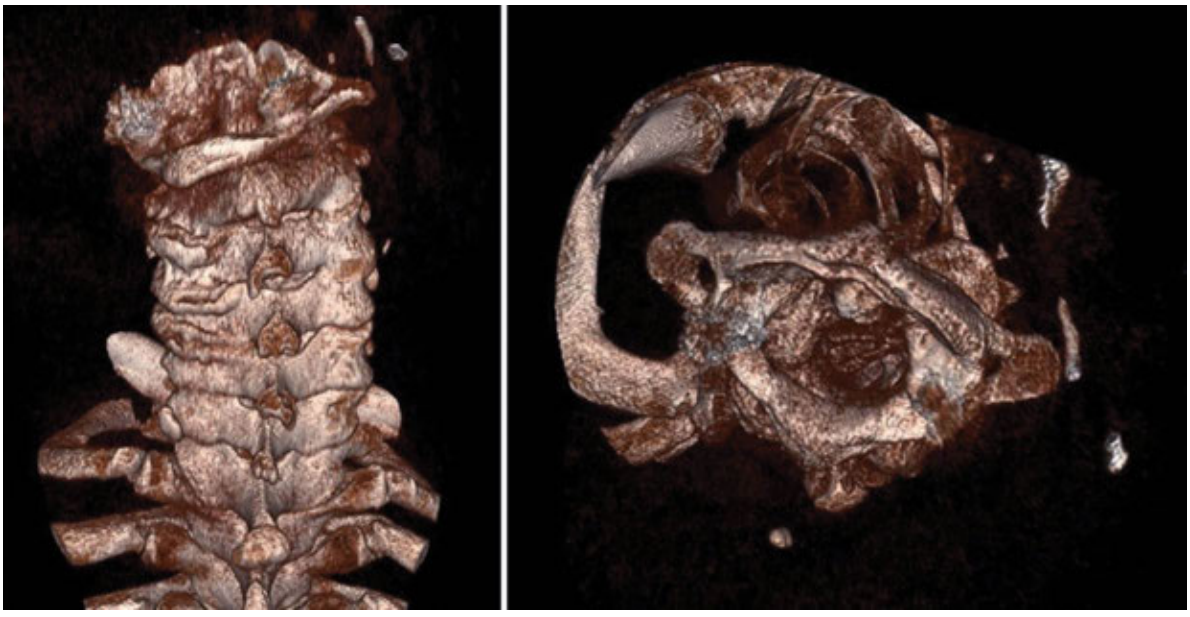

Figure 3. 3D CT reconstruction demonstrating Fielding Type I atlantoaxial rotatory fixation. The odontoid acts as a pivot point, with the left C1 facet subluxating anteriorly and the right C1 facet subluxating posteriorly. There is no anterior displacement.

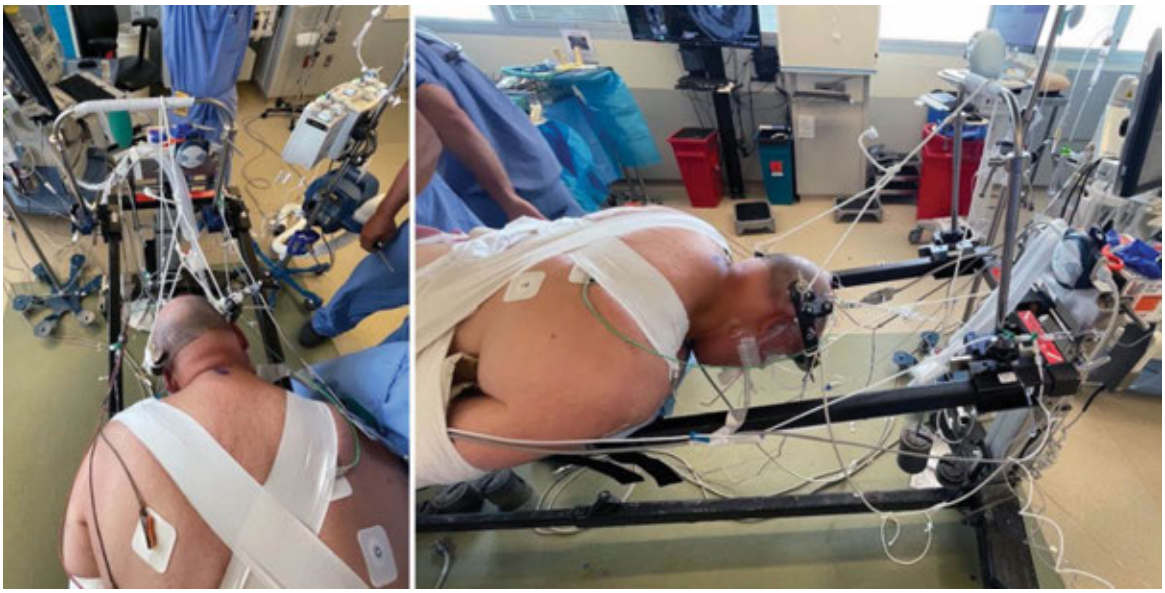

Figure 4. Intra-operative clinical photos showing the patient positioned prone on an open Jackson table with halo traction suspension applied.

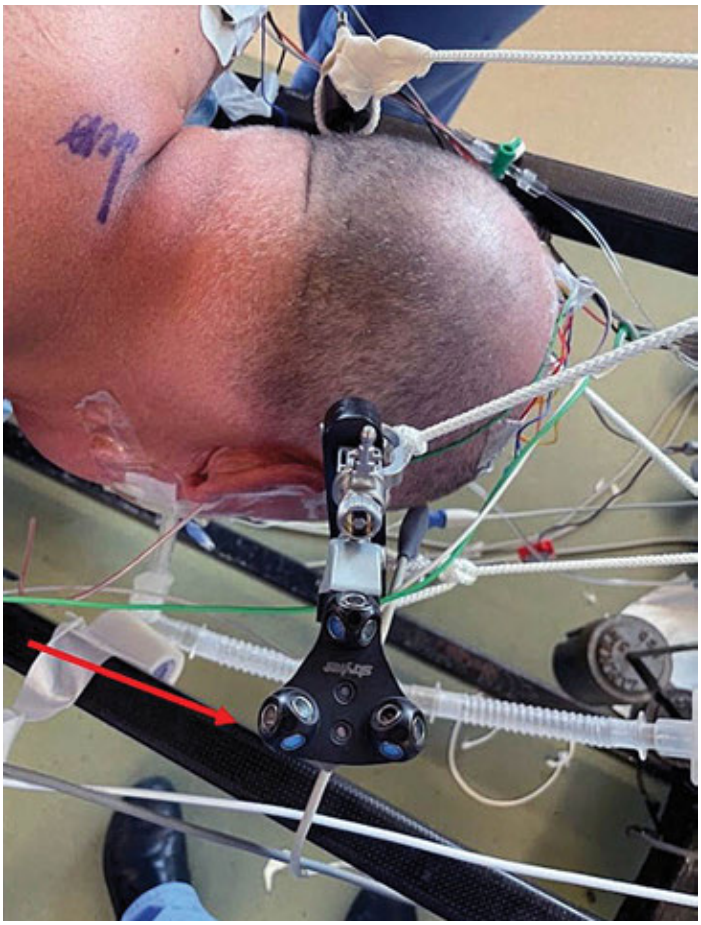

Figure 5. The navigation array shown attached to the halo (red arrow).

head to the right through the halo device. Reduction was visible along with an audible snapping sound. Fluoroscopic assessment confirmed appropriate alignment of the C1 and C2 vertebral bodies.

Unfortunately, the navigation array shifted during the reduction maneuver and the plan to navigate C1 and C2 screws was abandoned. Instead, fluoroscopic guidance was used for insertion of lateral mass screws into C1 and pedicle screws into C2. Rods were inserted. Distraction was applied on the left and compression on the right to further improve alignment of the cranium on the two superior vertebral bodies. A cross-link was applied at the caudal aspect of the construct to further limit rotation and prevent recurrent subluxation (Figure 6).

Next came bone grafting. After thorough decortication with a burr, iliac crest bone graft was harvested and laid between C1 and C2. The wound was then closed in a layered fashion. Drapes were taken down and a significant improvement in alignment was observed (Figure 7). The patient was re-positioned onto a hospital bed for removal of the halo and a hard cervical collar was applied. He was extubated in the operating room and transferred to the intensive care unit overnight for monitoring.

Post-Operative Course

Overnight the patient was stable and on post-operative day (POD) one, he was transferred to the floor. A CT scan was obtained, which confirmed safe placement of screws and appropriate reduction of the C1-C2 rotatory subluxation (Figure 8 & Figure 9).

Although the patient's alignment clinically was improved, he still had a slight persistent head yaw to the left (Figure 10). His left sternocleidomastoid contracture persisted. He elected to undergo left sternocleidomastoid tenotomy at the level of the clavicle on POD 3. He was then discharged home the following day on POD 4 in a hard cervical collar.

The patient was seen back in the clinic two weeks after discharge, at which time he reported 80% improvement in his symptoms (Figure 11). He remained in a hard cervical collar for ± weeks post-operatively. The patient returned to the office six months post-operatively and was satisfied with his alignment and range of motion.

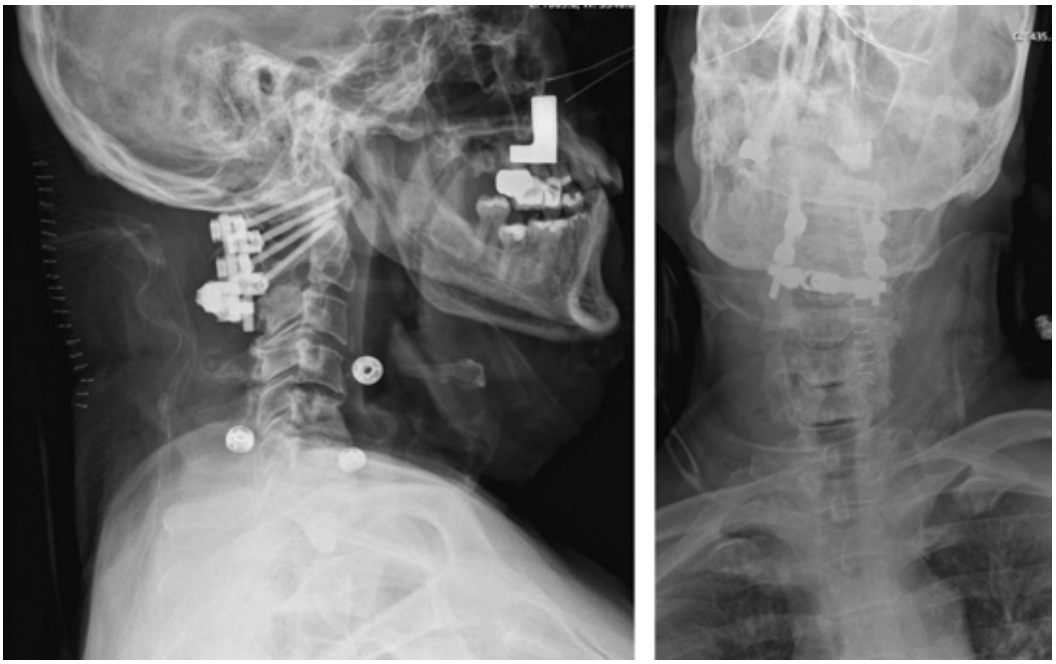

Figure 6. Lateral and AP radiographs showing the final construct, C1-C2 fusion with a cross-link at the caudal aspect of the construct.

Figure 7. Intra-operative comparison of head alignment prior to **(LEFT)** and at the completion of **(RIGHT)** surgery while still in halo traction suspension.

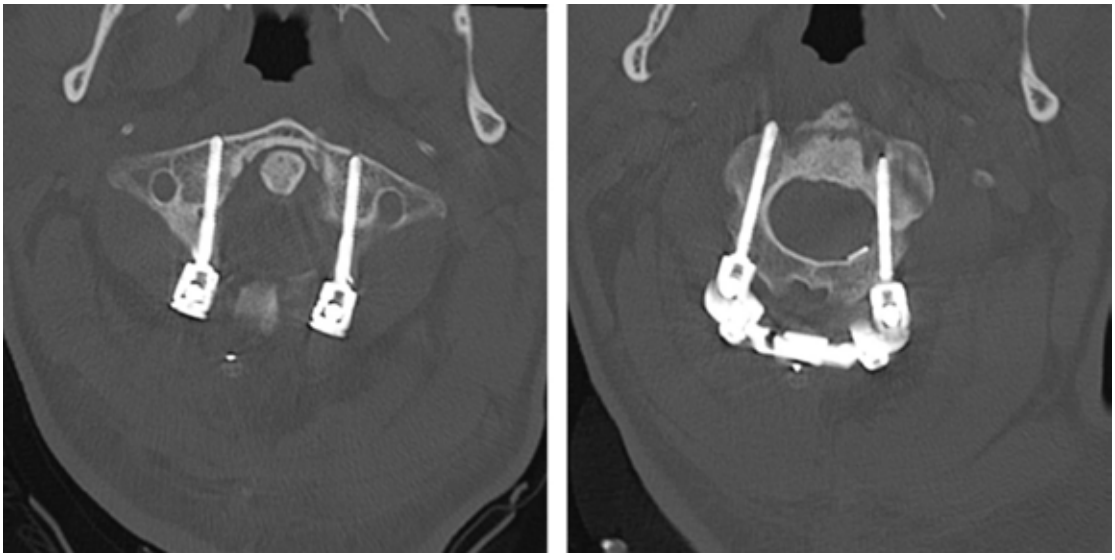

Figure 8. Post-operative axial CT images showing safe placement of C1 lateral mass screws **(LEFT)** and C2 pedicle screws **(RIGHT)**.

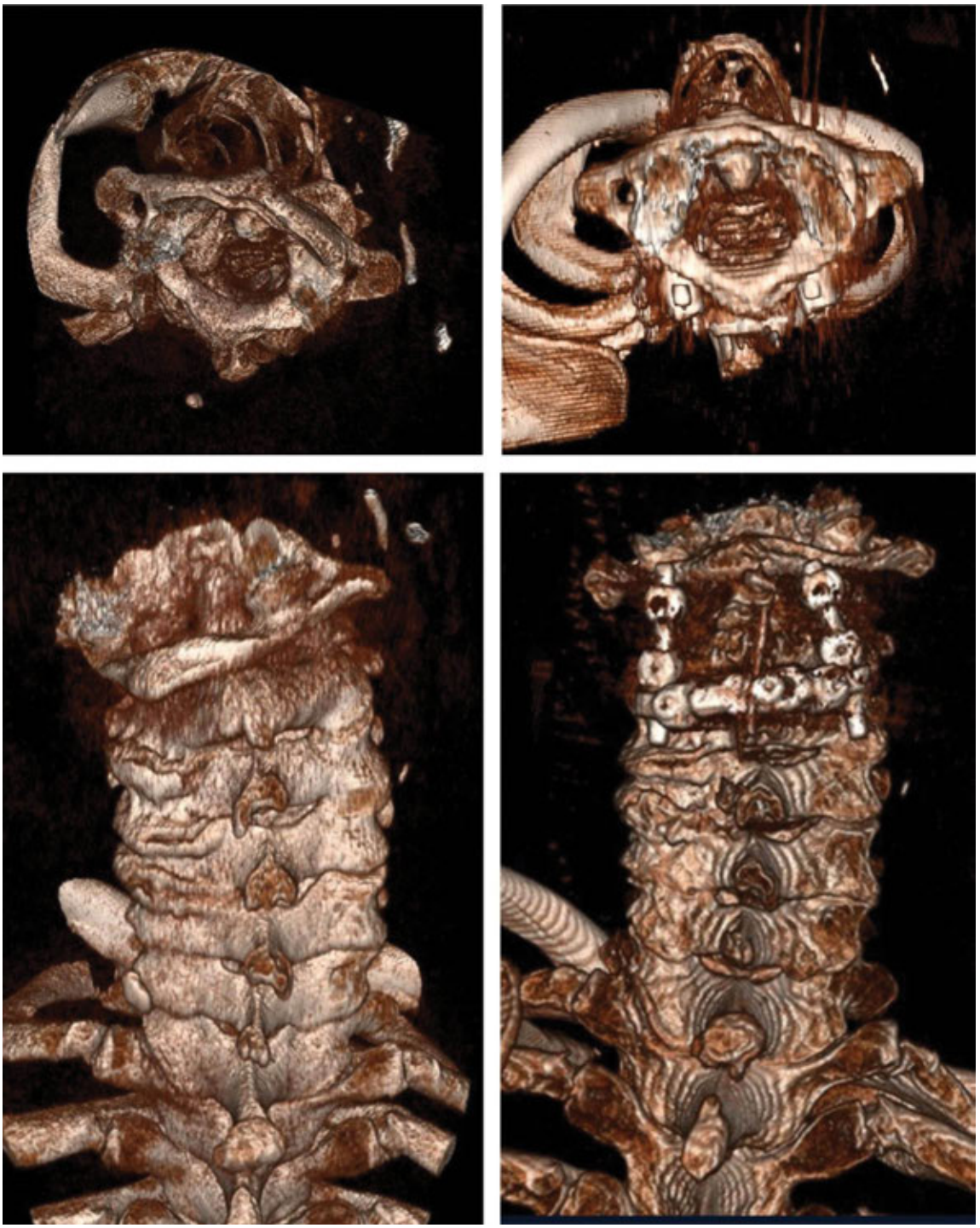

Figure 9. 3D CT reconstructions demonstrating interval reduction of C1-C2 rotatory subluxation (**LEFT:** Pre-Operative; **RIGHT:** Post-Operative).

Discussion

Atlantoaxial rotatory fixation (AARF) is a uncommon condition normally seen in the pediatric population and rarely seen in adults.[9,10] When it occurs in the adult population, AARF usually is caused by trauma, with case series citing automobile collisions, falls, and sport-related accidents as the most common etiologies.[10,11] Atraumatic AARF in adults, are even more rare, with only seven cases previously described in the literature.[1-7]

Regardless of the etiology, there is no consensus regarding the optimal strategy for managing this complex problem in adult patients.[12] Conservative treatment involves a trial of cranial traction with or without manipulation, generally followed by immobilization in a cervical collar or halo vest.[9,12] Additionally, there are varying expert opinions on the duration of conservative management.[12] In cases where no reduction is obtained with traction and manipulation, or in patients who experience re-dislocation, surgical treatment with C1-C2 fusion can be pursued.[9]

In the acute setting, conservative treatment has been successful.A recent literature review found that 25/31 patients who sustained traumatic AARF and initiated conservative

Figure 10. Clinical photos comparing head tilt before after the first stage of the procedure: **LEFT:** Head tilt pre-operatively before C1-C2 fusion.

RIGHT: Head tilt post-operatively after C1-C2 fusion, but prior to left sternocleidomastoid tenotomy.

Figure 11. Clinical photos at first post-operative visit, noting improvement in head alignment.

treatment within 1 month of injury successfully responded to traction and manipulation.[11]

Initially, AARF is often a missed diagnosis as patients can present with normal neurologic exams and non-diagnostic cervical spine radiographs, leading to delays in initiating conservative treatment.[13,14] For example, in a retrospective review of 26 patients treated for AARF at a single institution between 1988 and 2000, the authors found that it took an average of 15 months to establish the proper diagnosis of AARF.[9] Such a significant delay in diagnosis has major implications, as there is a correlation between time to diagnosis and failure of conservative treatment.[9,11] The same authors who found success with conservative treatment initiated within one month of dislocation also found that only 6/25 cases diagnosed after 1 month responded to conservative measures.[11] One possible explanation for this relationship is that muscle or ligamentous contractures may develop over time in response to the abnormal positioning of the C1-C2 joint.[11]

In these chronic cases where conservative treatment is unsuccessful or avoided entirely, open reduction with C1-C2 fusion can be performed. Multiple reduction strategies have been described, with several authors advocating for inserting temporary transverse rods at C1 and C2 as anchors to maneuver for reduction.[11,15]

Using a halo as the reduction tool, as in this case, has not previously been described in the literature. We feel this technique has multiple advantages relative to previously reported strategies. Use of a halo provides excellent rotational control of the head and allows manipulation of the device, making reduction maneuvers much easier. Compared to classic

head positioning with a Mayfield, the halo can be manipulated sterilely. When using halo traction suspension there is no need to have an assistant outside the sterile field to manipulate the Mayfield during attempted reduction maneuvers.

Although others have demonstrated success with the transverse rod technique described by Rajasekaran, the strategy relies on the strength of C1 screw purchase, which may not always be adequate to perform the reduction. This method place the C1 lateral arches at risk as the the screws can ploughing through them. Despite this theoretical risk, the transverse rod technique is an excellent adjunct if reduction maneuvers through the halo are not successful.

Conclusion

The diagnosis of atlantoaxial rotatory fixation is often missed in adults. Patients identified with the pathology early can be treated with nonoperative measures successfully. Delays in diagnosis are associated with failure of conservative treatment, and most patients with chronic AARF require posterior C1-C2 fusion.

Given the rarity of this condition in the adult population, controversy persists regarding therapeutic management, and multiple surgical techniques have been described. This case highlights the utility of halo traction suspension in achieving reduction of the C1-C2 joint in patients with chronic AARF.

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