Atlantoaxial Subluxation Secondary to SARS-CoV-2 Infection: A Rare Orthopedic Complication from COVID-19

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Patient: Male, 86-year-old

Final Diagnosis: Atlantoaxial subluxation

Symptoms: Neck pain

Medication: —

Clinical Procedure: —

Specialty: Rehabilitation

Objective: Rare coexistence of disease or pathology

Background: There are several reports of atlantoaxial subluxation caused by upper respiratory tract infections. Although, there are many known non-pulmonary complications COVID-19 infection, to date there have been no reported cases of orthopedic complications in the peer-reviewed literature. Diagnosis and management of atlantoaxial subluxation is currently limited. Therefore, it is important to explore other methods of identifying and treating patients suffering from atlantoaxial subluxation.

Case Report: Our patient was an 86-year-old man with right-sided neck pain and reduced range of neck motion for the past 6 months, shortly after a mild case of COVID-19. Autoimmune and inflammatory workup was unremarkable. Patient’s symptoms persisted despite 3 weeks of conservative therapy with analgesics, cervical collar, and physical therapy. He received low-frequency kinetically directed impulse wave (al-Kindi wave) treatment administered by the KKT device after 3-dimensional digital X-ray analysis of the atlas. After receiving the treatment over a period of 13 days, patient showed significant improvement in symptoms and follow-up X-rays.

Conclusions: For patient’s having neck stiffness or pain with COVID-19, it is important to consider atlantoaxial subluxation as a potential cause, especially if the patient requires intubation, as the technique should be adjusted to reduce spinal injury. Atlas X-ray analysis with Spinalytics provides very precise measurements of the atlas in relation to the skull and cervical spine, and improvement in angles were seen before and after treatment. The al-Kindi wave treatment was also effective in reducing the patient’s symptoms and improving cervical X-ray results, but further studies are required for confirmation.

Keywords: Joint Dislocations • COVID-19 • Neck Pain

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Background
As we continue to grapple with the SAR-CoV-2 pandemic, we are discovering numerous non-pulmonary complications of the virus. Studies have documented that the SARS-CoV2 virus impacts many other organ systems, such as the heart [1], bone marrow [2], kidney [3], liver [4], brain [5], and other organs [6]. However, no orthopedic complications have been reported.

In the adult population, the most common cause of non-traumatic atlantoaxial subluxation is rheumatoid arthritis [7]. However, there are also several reported cases of upper respiratory tract infections causing atlantoaxial subluxation in adults [8,9]. Currently, there are 2 mechanisms which help describe their association. The first theorizes that severe pharyngeal inflammation extends to the parapharyngeal space and then to the transverse ligament of the atlas, and the inflammation weakens the transverse ligament, causing subluxation. In the second mechanism, the infection enters the atlantoaxial joint through a hematogenous route [10]. Although COVID-19 also causes severe inflammation and accesses the bloodstream [11,12], no cases of atlantoaxial subluxation secondary to the COVID-19 virus have been described in the peer-reviewed literature.

In this case, we describe a healthy elderly man who had atlantoaxial subluxation shortly after a mild case of COVID-19. After initial conservative therapy was unsuccessful, the patient improved with wave treatment administered by the KKT device.

Case Report
The patient was an 86-year-old man with right-sided neck pain for the past 6 months. He also described stiffness and decreased range of motion in all directions, but particularly neck extension. He rated his neck pain as 9 out of 10 and noted that it worsened with movement in any direction. The pain and stiffness limited many of his activities, such as driving. The patient did not endorse a history of trauma, pre-existing bone disease, congenital disease, rheumatoid arthritis, or other autoimmune diseases.

Six months prior to presentation, the patient was diagnosed with mild COVID-19. He was treated symptomatically and fully recovered after 3 weeks of isolation. There was no evidence of bacterial co-infection and he improved without antibiotics. He did not require hospitalization or oxygen support. The patient did not have any post-infectious sequelae. He had a remote history of subtotal thyroidectomy performed 40 years ago. The patient had a 30-year history of diabetes mellitus type 2 and hypertension, for which he was taking glliclazide and amlodipine.

He was seen by a neurosurgeon 4 weeks prior for neck pain and stiffness and was advised to undergo conservative therapy with analgesics and a cervical collar. The patient did not improve after 3 weeks of follow-up, and he was advised to start physical therapy. Unfortunately, he was unable to tolerate physical therapy due to severe pain and was referred to the KKT Orthopedic Spine Center for wave treatment.

On examination, the patient’s head was tilted to the right side with left-sided torticollis (Figure 1A). There was a severe limitation in range of motion in all directions. Spasm of the SCM bilaterally was present, with some mild spams on the posterior regions of the neck. Palpation of the cervical spine (C1 to C6) produced pain. Generalized tenderness of the right posterior and lateral aspect of the neck was also noted. There was no evidence of joint tenderness or swelling in the fingers, wrists, elbows, shoulder, knees, or feet. The oral mucosa was also examined, and there was no pharyngeal erythema or enlarged tonsils. Vitals signs were within normal limits. Initial laboratory studies, provided in Table 1, were unremarkable.

Multiple cervical X-rays were taken in lateral and open-mouth views (Figure 2A-2C). The open-mouth view of the cervical radiograph revealed a right-tilted position of the atlas relative to the dens of the axis. The lateral view showed no thickening of the parapharyngeal soft tissue, and the distance between the axis and dens was within normal values for age (ADI <3.4 mm). This confirmed the diagnosis of Type 1 Rotatory Atlantoaxial Subluxation according to the Fielding and Hawkins classification.

Treatment
The KKT treatment delivers low-frequency kinetically directed impulses (al-Kindi waves) toward the atlas at frequencies of 16-200 Hz. The purpose of the treatment is to manage chronic pain. As part of the KKT treatment, patients require digital X-rays of the cervical spine from multiple directions (open-mouth, frontal, and lateral) for analysis by the Spinalytics software. The software describes the atlas position in relation to the skull and cervical spine to determine the appropriate vector to apply the al-Kindi waves to offset atlas deviation.

As shown in Figure 3A and 3B, the Spinalytics analysis showed a deviation of the atlanto-occipital angle 24.90° to the right (average 2.37° right), a clockwise rotation of the atlas of 6.95° in the frontal plane (average 1.86° clockwise), and a 5.00° rightward deviation of the atlanto-cervical angle (average 2.20° left). In the coronal plane, the atlas was rotated clockwise by 6.50° (average 2.14° counterclockwise). Average angles were obtained from an unpublished report calculating summary statistics of the Spinalytics database of patients with back and neck pain.
To offset the computed atlas deviation, the stylus of the KKT device was placed 1.5 cm below the tragus of the right ear. As shown in Figure 4A and 4B, the stylus was pointed at a height vector of 0.58 inches at 334°, with an impulse force of 1.5 lbs. The al-Kindi waves were applied in 18 treatment sessions over 13 days. Initially, the patient received 2 sessions daily, until it was gradually reduced to 1 session every 2 days.

Outcomes

After completing the treatment, the patient described significant improvement in pain (4/10) and increased range of motion in all directions, with the most significant improvement in neck extension. The right-sided tilt had also improved significantly. The patient noted considerable improvement in quality of life, with improvement in sleep, ability to drive, and performing activities of daily living. As shown in Figure 1B, the patient’s neck position improved appreciably.

Repeat X-rays were performed (Figure 2D-2F), which showed improvement in right-sided tilt in the open-mouth view. As shown in Figure 3C and 3D, the Spinalytics analysis showed a reduction of all the angles. The atlanto-occipital angle had shrunk from 24.90° to 11.70° to the right (average 2.37° right), and there was also a reduction in the clockwise rotation of the atlas from 6.95° to 4.15° in the frontal plane (average 1.86° clockwise). The rightward deviation of the atlanto-cervical angle decreased from 5.00° to 4.00° (average 2.20° left).

Table 1. Laboratory studies before treatment.

| Lab                      | Value     | Reference          |
|--------------------------|-----------|--------------------|
| White blood count        | 6.5×10^3/mL | 4.0-10.5×10^3/mL   |
| Hemoglobin               | 10.4 g/dL   | 14-18 g/dL         |
| RBC                      | 6.3×10^9/mL | 4.70-6.10×10^9/mL  |
| Hematocrit               | 45.2%      | 42.0-52.0%         |
| MCV                      | 90.2 fl    | 80-95 fl           |
| Platelet                 | 209×10^3/mL | 140-415×10^3/mL    |
| CRP                      | 6 mg/L     | <10 mg/L           |
| ESR                      | 13 mm/h    | 0-22 mm/h          |
| Rheumatoid factor (quantitative) | 10 IU/ml | <14 IU/ml          |
| Anti CCP Abs             | 0.5 U/ml   | 0-5 U/ml           |

Figure 1. (A) Image of the patient before treatment, showing the head’s right-sided deviation with torticollis. (B) Images of the same patient after treatment, showing normalization of head tilt.
coronal plane, the atlas clockwise rotation was reduced from 6.50° to 3.81° (average 2.14° counterclockwise).

**Discussion**

In this case study, the patient had considerable neck pain and stiffness after a recent case of COVID-19, which did not respond to conservative treatment. Ensuing cervical X-rays confirmed the diagnosis of rotatory atlantoaxial subluxation. The patient subsequently responded well to al-Kindi wave therapy administered by the KKT device.

There are 3 unique facets of this case that make it particularly interesting. 1) The patient presented with a previously unreported cause of atlantoaxial subluxation – COVID-19. 2) He underwent unique computerized imaging analysis to provide further insight about the atlas relative to the skull and cervical spine. 3) He improved after undergoing al-Kindi wave therapy.

**COVID-19 and Atlantoaxial Subluxation**

It is difficult to definitively attribute COVID-19 as the cause of atlantoaxial subluxation because there are other known causes that must first be ruled out. The primary differential diagnosis for causes of atlantoaxial subluxation in adults includes traumatic, infectious, inflammatory, and congenital causes [13]. This patient did not have an attributable history of trauma or congenital disease to implicate as the cause of his atlantoaxial subluxation. An inflammatory condition, such as rheumatoid arthritis, is also an improbable etiology, as the patient did not have peripheral joint symptoms or elevated serum markers.

Many bacterial and viral upper respiratory tract infections have been identified as triggers for atlantoaxial subluxation, either through a contiguous inflammatory process or by hematogenous spread [10]. However, there have been no documented cases describing COVID-19 as a possible source of atlantoaxial subluxation. In theory, COVID-19 may be a possible trigger, since its pathophysiology is similar to other known infectious
Figure 3. (A) Diagrammatic representation of the patient’s Spinalytics analysis, displaying a rotation of the atlas in posterior coronal view (red) with average angles (gray) before treatment. (B) Diagrammatic representation of the patient’s Spinalytics analysis displaying rotation of the atlas in inferior transverse view (red) with average angles (gray) before treatment. (C) Diagrammatic representation of the patient’s Spinalytics analysis, displaying a rotation of the atlas in posterior coronal view (red) with average angles (gray) after treatment. (D) Diagrammatic representation of the patient’s Spinalytics analysis displaying rotation of the atlas in inferior transverse view (red) with average angles (gray) after treatment. Images are for diagrammatic purposes only, and angles and distances are not accurately illustrated.
causes of atlantoaxial subluxation. This is the first reported case providing evidence for such a premise.

It is essential to recognize this association, as it may have implications in the inpatient setting. Approximately 80% of critically ill COVID-19 patients require intubation and mechanical ventilation during their care [14]. Due to cervical instability, atlantoaxial subluxation is a predictor of difficult intubation [15]. Therefore, practitioners must adjust the position and technique of intubation, with studies showing a benefit of the protrusion position, to provide more support and extension at the craniovertebral junction [16]. Although further inquiry is required, it may be prudent to use such a technique with COVID-19 patients who report neck pain or stiffness.

Computerized Imaging Analysis

The Spinalytics analysis described in this case study provides very detailed and precise information regarding the rotation and position of the atlas relative to nearby structures. The images are taken on a standardized track with head clamps to ensure consistent head placement to generate reproducible images that can be digitally analyzed. Previous studies have shown this methodology limits inter- and intra-observer variability [17]. As shown in Figure 3A, our patient had greater atlas rotation that average. After receiving treatment, a significant reduction in all the angles was noted.

It is estimated that 40-85% of rheumatoid arthritis patients with neck pain have atlas instability [18], and approximately 10% die of undiagnosed spinal cord or brain stem compression [19]. Since there are preventative procedures available [20], there is a role for the development of more accurate diagnostic tools for the early detection of at-risk patients. Further studies are required to determine if such detailed information can guide treatment or provide prognostic information.

Al-Kindi Wave Therapy for Atlantoaxial Subluxation

Unless there is evidence of neurovascular compromise, severe cervical instability, or unremitting pain, atlantoaxial subluxation is treated non-surgically [21]. In our case, the patient had a Type 1 Fielding and Hawkins subluxation without neurovascular symptoms. Most atlantoaxial subluxations secondary to infection improve spontaneously [22]; however, this was not the case for our patient. A subsequent trial of conservative therapy also did not improve his symptoms.

After 6 months of failed conservative therapy, the patient underwent al-Kindi wave treatment administered using the KKT device. The treatment applies impulses directed toward the atlas to offset deviations detected on digitally analyzed X-ray films. Previous studies have shown that the therapy can improve neck pain and restore cervical spine alignment [23,24]. This case shows that after receiving the al-Kindi wave therapy, there was significant improvement in symptoms and functional status, which was corroborated by follow-up imaging and digital analysis of the atlas.

Considering the KKT treatment aims to correct atlas deviation, the KKT treatment may be a rational treatment for reversing atlantoaxial subluxation. In particular, it may be helpful for patients who failed conservative therapy and would like to avoid surgery. Further studies are required to better...
understand its efficacy and role in the management of atlantoaxial subluxation.

Conclusions

For COVID-19 patients with neck stiffness or pain, it is important to consider atlantoaxial subluxation as a potential cause, especially if intubation is required, as the technique should be adjusted to reduce spinal injury. Atlas X-ray analysis with Spinalytics provides very precise measurements of the atlas in relation to the skull and cervical spine, and improvement in angles were seen before and after treatment. However, further studies are required to determine the clinical significance of this information. The al-Kindi wave treatment was also effective in reducing this patient’s symptoms and cervical X-rays, but further studies are required to determine if it could be a viable treatment for such patients.

Declaration of Figures’ Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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