Conservative management of idiopathic anterior atlantoaxial subluxation without neurological deficits in an 83-year-old female: A case report

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Atlantoaxial subluxation that is not related to traumatic, congenital, or rheumatological conditions is rare and can be a diagnostic challenge. This case report details a case of anterior atlantoaxial subluxation in an 83-year-old female without history of trauma, congenital, or rheumatological conditions. She presented to the chiropractor with insidious neck pain and headaches, without neurological deficits. Radiographs revealed a widened atlantodental space (measuring 6 mm) indicating anterior atlantoaxial subluxation and potential sagittal atlantoaxial instability. Prompt detection and appropriate conservative management resulted in favourable long-term outcome at 13-months follow-up. Conservative management included education, mobilizations, soft tissue therapy, monitoring for neurological progression, and co-management with the family physician. The purpose of this case report is to heighten awareness of the clinical presentation of idiopathic anterior atlantoaxial subluxation without neurological deficits. Discussion will focus on the incidence, mechanism, clinical presentation, and

La subluxation atloïdo axoïdienne qui n'est pas liée à des conditions traumatiques, congénitales ou rhumatologiques est rare et peut présenter un défi sur le plan du diagnostic. Cette étude de cas décrit un cas de subluxation atloïdo axoïdienne antérieure chez une femme de 83 ans sans antécédents de pathologies traumatiques, congénitales ou rhumatismales. Elle s'est présentée chez le chiropraticien avec des douleurs cervicales insidieuses et des maux de tête, sans déficits neurologiques. Les radiographies ont révélé un espace atlanto-dental élargi (6 mm) indiquant une subluxation atloïdo axoïdienne antérieure et la possibilité d'une instabilité atloïdo axoïdienne sagittale. La détection rapide et un traitement conservateur approprié ont mené à un résultat favorable à long terme, avec un suivi après 13 mois. Le traitement conservateur comprend la sensibilisation, les mobilisations, le traitement des tissus mous, le suivi de la progression neurologique, et la cogestion avec le médecin de famille. Cette étude de cas vise à la sensibilisation de la présentation clinique d'une subluxation atloïdo axoïdienne antérieure idiopathique sans déficits neurologiques. La discussion portera sur l'incidence, le mécanisme, la présentation clinique

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conservative management of a complex case of anterior atlantoaxial subluxation.

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et le traitement conservateur d'un cas complexe de subluxation atloïdo axoïdienne antérieure.

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MOTS CLÉS : subluxation atloïdo axoïdienne, cervicale supérieure, non traumatique, idiopathique, traitement conservateur

Introduction

Anterior atlantoaxial subluxation is characterized by a radiographic distance of more than 3 mm between the anterior aspect of the dens and the posterior aspect of the anterior arch of the atlas.1 The atlantoaxial subluxation can be considered stable/fixed or unstable/dynamic, which is often assessed with flexion and extension cervical radiographs. Stable or fixed atlantoaxial subluxation can involve significant widening of the joint space that does not change between the two views. Conversely, unstable anterior atlantoaxial subluxation is diagnosed when the anterior atlantoaxial diameter differs ≥ 2 mm between flexion and extension radiographs.1 Unstable anterior atlantoaxial subluxation, a form of atlantoaxial instability, can result in compression of the spinal cord or vertebral arteries.² Common causes of atlantoaxial instability are rheumatological (e.g. rheumatoid arthritis), congenital (e.g. Down Syndrome, os odontoideum), and traumatic conditions (e.g. dens fracture).³⁻⁷ Atlantoaxial instability that is not related to any predisposing condition is rare and can be considered idiopathic.

The clinical presentation of atlantoaxial instability varies widely. Severe cases can present with progressive myelopathy, vertebrobasilar insufficiency, or quadriplegia.⁸ Less severe signs and symptoms include neck pain, apprehension with neck movement, headaches, intolerance to prolonged static posture, and increased muscle tone.⁹⁻¹³ Mild cases can present without neurological deficits and mimic the clinical presentation of mechanical neck pain or cervicogenic headaches. It can therefore be difficult to detect atlantoaxial instability in these patients, particularly in those without associated rheumatological, congenital or traumatic conditions.

Although atlantoaxial instability may be a diagnostic challenge, timely diagnosis is needed to facilitate import-

ant considerations for its management. Surgical fusion is often required for severe or progressive neurological deficits, or vertical translocation.¹⁴ Conservative management may be considered for those with surgical indicators. However, there is a reported paucity of high quality literature on conservative management for mild cases without neurological deficits.¹⁴ This makes it difficult for primary contact providers and patients to make decisions around the management of atlantoaxial instability in mild cases without neurological compromise.

This case report details anterior atlantoaxial subluxation in an 83-year-old patient with no history of trauma, rheumatological, or congenital conditions. It was a diagnostic challenge, as the patient presented to the chiropractor with neck pain and headaches, no neurological deficits, and subtle non-mechanical symptoms. Subsequent radiographs revealed a widenened atlantodental space that measured 6 mm, indicating anterior atlantoaxial subluxation and potential sagittal atlantoaxial instability. The patient achieved favourable long-term outcome with conservative management by the chiropractor and comanagement with the family physician. The incidence, mechanism, clinical presentation, treatment considerations, and limitations of this case report will be discussed.

Case Report

An 83 year-old retired female presented to a chiropractic clinic with bilateral neck pain and headaches that were worse on the right. The pain started eight weeks ago of insidious onset with an intensity that fluctuated during the day. At the time of the encounter, the pain intensity was rated a 5/10 on a verbal numeric pain rating scale. The neck pain was felt in the bilateral suboccipital and trapezius region, while the headaches were felt in the bilateral temporal region and vertex of the head. The neck pain pre-

viously traveled down her right lateral arm, but was now minimal. Aggravating factors included neck extension for both neck pain and headaches. Coughing or straining did not aggravate the headaches. Self-administered massage was relieving, and the pain had improved slightly over time. There were no associated symptoms or red flags. The patient felt the neck pain and headaches when moving in bed, but they were not interfering with her sleep.

Her medical history was remarkable for high blood pressure and high cholesterol, which were being controlled by medications (i.e. Amlodipine and Lovastatin). Her health was otherwise unremarkable, though her family history was unknown. The patient did not smoke or drink alcohol and would perform simple stretches at home on a daily basis. She previously worked as a restaurant owner but had retired. No previous neck pain, headaches, allergies, trauma, or surgeries were reported. Systems review was unremarkable. The patient saw her family physician three weeks ago for this complaint and was provided education and reassurance that the pain would resolve.

On examination, the patient was 125 lbs and 5'1", blood pressure was 150/100, and all other vital signs were unremarkable. Mild anterior head carriage, rounding of shoulders, and a small bruise on the right lateral arm (attributed to self-massage) was noted. Cervical motion was full in flexion but produced mild right neck pain. Cervical motion was decreased by 50% in all other directions, and produced neck pain on extension and bilateral lateral flexion. Right Kemp's (i.e. passive ispilateral rotation, extension, and lateral flexion of the cervical spine) was positive for neck pain, while orthopedic tests for nerve root irritation and compression were negative. Palpation revealed tight and tender sternocleidomastoids, suboccipitals, trapezius and levator scapulae bilaterally, worse on the right.

Upper limb neurological examination was bilaterally present and symmetric for sensation and 5/5 for motor strength bilaterally. Deep tendon reflexes were 1+ for right biceps and 2+ for left biceps, 1+ bilaterally for brachioradialis, and 0 bilaterally for triceps. Hoffman's (i.e. flicking of the distal end of the third finger in flexion) and cranial nerve screen were unremarkable. The headache could not be reproduced during the examination, but the patient felt the headache at the vertex of her head immediately after the examination was completed.

The chiropractor suspected a resolving right C5 radiculopathy, cervicogenic headaches and bilateral mechanical neck pain. For symptomatic relief, a trial of treatment for 3-4 weeks consisting of education, soft tissue therapy, cervical and upper thoracic joint low-velocity, low-amplitude mobilizations, and exercises was recommended. High-velocity, low-amplitude manipulation of the cervical spine was not recommended at this time. The chiropractor advised monitoring for neurological progression, and a visit to the family physician to reassess her blood pressure and medication use. The chiropractor sent a letter to the family physician outlining the examination results and plan of management.

The patient had five chiropractic treatments over three weeks. During this time, the patient saw her family physician, who increased her dose of hypertensive medication. On re-evaluation with the chiropractor, the patient noted 50% improvement in the intensity and frequency of symptoms. She now experienced the neck pain and headaches in the morning and when lying down, and they improved towards the evening. She described her head as feeling heavy upon waking in the morning, which lasted a few minutes. Although the patient reported improvement, the non-mechanical symptoms warranted further investigation and the chiropractor ordered cervical radiographs.

The cervical radiographs (Figures 1A, 1B, and 1C) revealed: 1) atlantodental space measured 6 mm, indicating anterior atlantoaxial subluxation and potential sagittal atlantoaxial instability, with borderline spinal stenosis at C1; 2) degenerative disc disease from C3-7 with central stenosis at C4 and C5; 3) moderate uncovertebral arthrosis at C3-4, C4-5, C5-6 and C6-7; 4) postural alterations; 5) moderate osteopenia; and 6) arteriosclerosis of aortic knob. The radiologist recommended a neurological examination for cervical spondylotic myelopathy and an orthopedic consultation.

The chiropractor recommended a visit to the family physician, suggesting flexion/extension views to assess whether the anterior atlantoaxial subluxation was unstable and an orthopedic referral. The chiropractor also performed a lower limb neurological examination, which was unremarkable. Subsequently, the family physician suggested continuing with chiropractic treatment given the improvement, and monitoring for any neurological progression. The family physician decided that flexion/ extension radiographs and an orthopedic referral may be considered later if the patient did not improve with treatment. The patient's blood pressure was 130/100, and was



Figure 1A: Lateral Cervical Radiograph Bone density is moderately diminished, but vertebral body heights were well maintained and no congenital bony anomaly was visualized. Cervical lordosis is mildly reversed with a large atlantodental space measuring 6 mm. The space available for spinal cord (SAC) measures 15 mm at C1. Disc narrowing with bone spurring is noted from C3-7. The sagittal spinal canal is narrowed measuring 12 mm at C4 and 9 mm at C5. The facet joints and prevertebral soft tissue are unremarkable.



Figure 1B: Anterior Posterior Open Mouth Cervical Radiograph Mild joint space narrowing is noted in the right C1-2 articulation.



Figure 1C: Anterior Posterior Lower Cervical Radiograph Uncinate blunting is noted at C4, C5, C6 and C7. The facet joints are unremarkable. Calcific plaques are noted at the aortic knob. being controlled by medication. However, the patient still experienced neck pain and headaches at the time, and decided to continue with chiropractic treatments.

The patient continued with four chiropractic treatments over six weeks, with emphasis on neck strengthening exercises. The cervical joint mobilizations were now performed only in a neutral cervical spine position, and included shearing of the cervical joints segmentally in lateral flexion and extension. Soft tissue therapy to tight neck muscles and home exercises were continued. The neck strengthening exercises consisted of isometric exercises in flexion, extension and bilateral lateral flexion. Gentle exercises such as shoulder rolls and chin tucks were also prescribed for the patient. Emphasis was given to perform these exercises in the morning when her head felt heavy, since she noted relief with exercises.

On re-evaluation after six weeks by the chiropractor, the patient reported 90% resolution of the headache and neck pain, and no difficulty performing normal activities. The patient was able to self-manage with daily exercises and no longer had any pain or restricted cervical motion. Minimal pain was produced on palpation of the suboccipital muscles, but the examination was otherwise unremarkable. The patient was discharged from treatment and was informed to return to the family physician and chiropractor if symptoms regressed.

Two months later, a follow-up phone call made by the chiropractor found that the patient no longer had any symptoms and was still performing her neck strengthening exercises daily. The patient maintained complete resolution of symptoms at 13 months follow-up and was satisfied with her chiropractic care.

Discussion

Incidence:

Atlantoaxial subluxation involves a widened atlantoaxial joint that can be stable or unstable with movement.¹ Unstable or dynamic anterior atlantoaxial subluxation (i.e. with instability) is characterized by a difference of at least 2 mm in the anterior atlantoaxial diameter between flexion and extension radiographs.¹ Atlantoaxial instability is most commonly reported in patients with trauma or preexisting conditions such as rheumatoid arthritis or Down syndrome. Up to 50% of patients with rheumatoid arthritis for more than seven years report atlantoaxial instabiliity.³ Atlantoaxial instability affects 10-20% of individuals with Down Syndrome.⁴ Traumatic atlantoaxial instability reportedly occurs in approximately 35% of cases with type II odontoid fractures, and less than 5% of cervical trauma cases without concomitant fractures.^{15, 16} Atlanto-axial instability in absence of trauma or these pre-existing conditions appears rare and has been reported in only two case reports and one retrospective case series.¹⁷⁻¹⁹ To our best knowledge, no data exists regarding the prevalence of atlantoaxial instability without predisposing factors in the general population.

Mechanism:

The pathogenesis of atlantoaxial instability in certain traumatic, rheumatological, and congenital conditions has been described in previous literature. In rheumatoid arthritis, the instability is secondary to destruction of articular and ligamentous structures from chronic synovial inflammation.²⁰ In Down Syndrome, the phenotypic feature of generalized ligamentous laxity is responsible for craniocervical instability and dislocation.²¹ The development of atlantoaxial instability after traumatic conditions is the result of a fracture of the atlas or odontoid process, and/or traumatic rupture of transverse or alar ligaments.⁶

However, the etiology of atlantoaxial instability in absence of these conditions is not well understood. The few studies describing this occurrence attributed osteoarthritis at the C1-C2 joints in the elderly population as the cause of atlantoaxial instability.^{18,19,22} A retrospective analysis of 108 subjects with atlantoaxial instability and moderate-to-severe degeneration at C1-C2 joints found that only 40% of cases had head or neck trauma.¹⁹ The remaining cases had no known underlying conditions that could have resulted in atlantoaxial instability. The authors suggested that degenerative hypertrophy and weakening of periodontoid and periarticular ligaments may have contributed to the instability in these cases.¹⁹ However, it is important to note that case reports and retrospective analyses are unable to determine causal relationships.

Our patient had only mild degeneration at the C1-C2 articulation. Our patient also did not have any preceding trauma or underlying rheumatological or congenital conditions that could have been attributed to the atlantoaxial instability. It can be noted that she did have moderate-tosevere degeneration in the lower cervical spine. However, cervical degeneration was likely an unrelated finding to her neck pain and headaches. The Bone and Joint Decade 2000-2010 Task Force on Neck Pain and Its Associated Disorders found no admissible evidence suggesting that cervical degeneration was a risk factor or prognostic factor for neck pain and associated disorders.^{23,24} Our case report sheds light on the potential for symptoms related to atlantoaxial instability in the absence of severe degeneration at the C1-C2 joints, though its mechanism remains unclear.

Clinical Presentation:

The clinical presentation of atlantoaxial instability can be highly variable and lead to diagnostic challenges. Some cases are reported to be asymptomatic until the spinal cord is irreversibly compromised at a posterior atlantodental interval of 14 mm or less.²⁵ Signs and symptoms can include occipital neuralgia (through compression of the greater occipital nerve), headaches, neck pain, loss of cervical motion, and progressive sensory and motor dysfunction in the upper and lower extremities.^{18,19,26} More severe clinical presentation includes radiculopathy, myelopathy, quadriplegia, and, in extreme instances, sudden death.⁸

The clinical presentation of atlantoaxial instability in patients without trauma or predisposing conditions appears to be milder or have slowly progressive neurological deficits. A report of two cases described subjects with idiopathic atlantoaxial instability (measured as 8 mm atlantodental interval) who presented with cervico-occipital pain and no neurological deficits.¹⁸ One subject reported radiation of pain to the right parietal and occipital aspects of the scalp, upper trapezius, and shoulder. Another subject reported radiation of pain to the left parieto-occipital and retro-auricular areas. Both subjects had decreased cervical motion in all directions except flexion and extension and had moderate C1-C2 degeneration on radiographs.¹⁸ In the retrospective analysis of 108 subjects with craniovertebral instability attributed to upper cervical degeneration, all subjects presented with neck pain, restricted cervical motion, and cervical muscle spasm.¹⁹ These subjects also presented with sensory deficits that were mild and slowly progressive.19

Our patient experienced symptoms similar to the milder clinical presentation reported in the literature. Recent onset of headache and bilateral neck pain characterized our patient's chief complaint. The patient also reported subtle non-mechanical patterns of pain, including aggravation of pain when lying down and resting in bed, and improvement of pain during the day. The feeling of heaviness of the head was also atypical of mechanical neck pain and headaches. It is important for primary contact providers, including chiropractors, to be aware of the clinical picture of atlantoaxial instability. A retrospective review of 847 chiropractic patient charts with radiographs found 0.6% of patients had atlantoaxial instability.²⁷ Therefore, this condition, though rare, may present to chiropractic clinics. An appropriate clinical index of suspicion for underlying pathology prompted the chiropractor in this case to investigate further with radiographs.

Management:

Generally accepted indications for surgical intervention include intractable pain, severe neurologic deficits, or vertical translocation with compromise of the vertebral artery.¹⁴ Moreover, surgical indicators for atlantoaxial instability related to rheumatoid arthritis include atlantoaxial impaction, cord stenosis, and intractable pain unresponsive to conservative treatment that is affecting daily activities.^{20,28} In a retrospective analysis of 108 cases with slowly progressive sensory deficits from idiopathic atlantoaxial instability, all cases received surgical fusion.¹⁹ This suggests that the presence of progressive neurological deficits may also be an indication for surgery.

The decision between conservative and surgical management for mild cases of atlantoaxial instability without neurological deficits is less clear. Conservative management has been suggested for mild complaints and to achieve temporary relief for neurological deficits.²⁶ However, there is a paucity of high quality studies examining the effectiveness of conservative versus surgical management for atlantoaxial instability. A systematic review by Wolfs et al assessed the neurologic outcome and survival time of patients with rheumatoid cervical spine subluxation after surgical or conservative treatment.14 The systematic review found that neurologic outcomes after surgery were superior to conservative treatment in all patients with some neurological deficits. However, surgical and conservative management yielded similar outcomes in patients who had no neurological deficits. All included studies had high risks of bias, and generalizability was limited to those with rheumatoid arthritis.¹⁴ This suggests, in part, that conservative management can be considered in patients without neurological deficits from atlantoaxial instability of an idiopathic origin.

Little is known about which interventions should be used in conservative management of atlantoaxial instability. Kauppi et al detailed the use of a custom made stiff collar in a small case series of patients with unstable anterior atlantoaxial subluxation due to rheumatoid arthritis.²⁹ It was suggested that a collar can be used in mild forms of atlantoaxial instability, particularly for those without atlantoaxial subluxation in the neutral position.²⁹ Kauppi et al also investigated a course of multidisciplinary treatment (given by a rheumatologist and physiotherapist) for adult patients with rheumatoid atlantoaxial instability.³⁰ The treatment involved education, isometric neck exercises, relaxation exercises, massage, collars, ergonomics, and active disease-modifying medication. Cervical pain was substantially reduced post-intervention and this was maintained at 12 months.³⁰ Specific to idiopathic atlantoaxial instability, one case report documents the use of medication and a soft collar, while another documents the outcomes of no intervention.¹⁸ Both cases reported stable clinical and neurological presentation at 18 months follow-up.¹⁸ It is not clear which conservative intervention is superior to one another based on the current literature.

In our case, the decisions around conservative management were based on clinical reasoning and limited literature in this area. The chiropractor aimed to provide soft tissue therapy for tight musculature, joint mobilizations in neutral spine positions for pain relief, and strengthening exercises for stabilization. The chiropractor avoided using end range positions and spinal manipulation to be cautious, particularly with unknown etiology for the patient's atlantoaxial instability. Conditions involving ligamentous laxity and potential anatomic subluxation or dislocation have been reported as absolute contraindications to high-velocity thrust procedures in anatomical regions of involvement.³¹ These conservative interventions resulted in favourable long term outcome for this patient at 13 months follow-up, and may be studied further in future research.

Limitations:

There are limitations to this case report. The anterior atlantoaxial subluxation was not further assessed with flexion/extension radiographs to determine if atlantoaxial instability was present. It was unknown whether the patient

suffered from a stable or unstable anterior atlantoaxial subluxation. Therefore, the results of this case report are specific to anterior atlantoaxial subluxation, and may differ from cases with atlantoaxial instability. There were also other variables that may have led to the patient's headaches. First, the patient may have been experiencing hypertensive headaches, since she had elevated blood pressure when she first presented to the chiropractor. However, this is not likely the case for a number of reasons. The patient had previous episodes of elevated blood pressure, but this was the first time she experienced these headaches and neck pain. The headaches and neck pain also remained after the blood pressure decreased with a new dosage of antihypertensive medication from the family physician. The headaches were not aggravated by coughing or sneezing, which is one of the criteria listed for hypertensive headaches by the International Classfication of Headache Disorders second edition.32

Moreover, current literature suggests that individuals with higher systolic blood pressure (unless above 180) were up to 40 percent less likely to have headaches compared to those with healthier blood pressure readings.³³ The patient's blood pressure was 150/100 and therefore not associated with an increased likelihood for headaches. Second, the patient may have been experiencing headaches associated with neck pain, such as cervicogenic headaches. However, her headaches presented in a non-mechanical nature, so this was unlikely the case. For instance, the headaches were worse in the morning and with lying down, and improved towards the evening. Moreover, the headaches could not be reproduced during physical examination or with palpation of the neck.

Lastly, the patient had multilevel uncovertebral osteoarthrosis from C3 to C7. In 2008, the Bone and Joint Decade 2000-2010 Task Force on Neck Pain (NPTF) found three scientifically admissible studies examining the relationship between degeneration and neck pain prevalence.³⁴ Two of the studies did not find any significant difference between degree of neck pain and radiographic evidence of cervical spine degeneration in females.^{35,36} One study showed increasing prevalence of neck pain with increasing grade of atlanto-odontoid osteoarthrosis.³⁷ However, the patient in our case had only mild joint space narrowing in the right C1-2 articulation. Moreover, the NPTF did not find any admissible studies examining the role of degenerative changes as a prognostic factor for neck pain in the general population.²⁴ Future studies with confirmed atlantoaxial instability are needed to examine the effectiveness of conservative interventions for cases with no or minimal neurological deficits. Overall, this case report highlights the critical thinking process involved in managing a complex case of suspected atlantoaxial instability without neurological deficits in an elderly patient.

Summary:

Atlantoaxial subluxation of idiopathic origin is rare and can be challenging to diagnose or manage clinically. This case report highlighted the detection of idiopathic anterior atlantoaxial subluxation and potential sagittal atlantoaxial instability without neurological deficits in an elderly female. Favourable long term outcome was achieved with conservative management. The case helps to heighten awareness of the clinical presentation and treatment considerations around idiopathic anterior atlantoaxial subluxation among primary contact providers.

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