Symptomatic improvement in function and disease activity in a patient with ankylosing spondylitis utilizing a course of chiropractic therapy: a prospective case study

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Background: There is limited outcome measure support for chiropractic manipulative therapy in the management of ankylosing spondylitis. An improvement in specific indices for both function and disease activity during chiropractic therapy for ankylosing spondylitis has not previously been reported.

Objective: To measure changes in function and disease activity in a patient with ankylosing spondylitis during a course of chiropractic therapy. The clinical management of ankylosing spondylitis, including chiropractic manipulative therapy and the implications of this case study are discussed.

Clinical Features: A 34-year-old male with a 10 year diagnosis of ankylosing spondylitis sought chiropractic treatment for spinal pain and stiffness. His advanced radiographic signs included an increased atlantodental interspace and cervical vertebral ankylosis.

Intervention and outcome: The Bath Ankylosing Spondylitis Functional Index (BASFI), Bath Ankylosing Spondylitis Disease Activity Index (BASDAI), finger-tip-to-floor distance and chest expansion were assessed during an 18 week course of chiropractic spinal manipulation and mobilization therapy. There was a 90% improvement in the disease activity index and an 85% improvement in the functional index from the pre-treatment baseline, as measured by the BASDAI and BASFI respectively. Spinal flexibility and chest expansion also improved.

Conclusion: To the authors knowledge this is the first study to incorporate ankylosing spondylitis specific
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indices, for both disease activity and function, to objectively support the use of chiropractic manipulative therapy in the management of ankylosing spondylitis. More intensive research is suggested.

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KEY WORDS: ankylosing spondylitis, chiropractic, manipulation, Bath Ankylosing Spondylitis Functional Index (BASFI), Bath Ankylosing Spondylitis Disease Activity Index (BASDAI).

Introduction

We report the case of a 34-year-old male who experienced symptomatic improvement in function and disease activity during chiropractic therapy for ankylosing spondylitis (AS). An objective improvement in both function and disease activity has not previously been reported in a patient with ankylosing spondylitis utilizing chiropractic manipulative therapy. The clinical management of ankylosing spondylitis and the implications of this case study for the use of chiropractic manipulative therapy is considered.

Ankylosing spondylitis is a chronic and progressive spondyloarthropathy most commonly affecting the axial skeleton of young males. It is characterized by articular bony ankylosis, ligamentous ossification and enthesopathies. The most common sites of involvement are the sacroiliac, apophyseal, costovertebral, pubic symphysis, discovertebral and manubriosternal joints. In the spine, AS is localized to the discovertebral junction, apophyseal, costovertebral joints and the atlantoaxial joint. The pathogenetic radiographic sequence of AS at the discovertebral junction begins with the Romanus lesion, which creates the vertebral body squaring associated with AS. Healing of the lesion leads to refractive sclerosis or the shiny corner sign. Further progression of the disease leads to ossification of the spinal ligamentous tissues which result in marginal syndesmophytes. Sacroiliac joint involvement is the hallmark of AS and is most commonly bilateral and symmetric in presentation. AS is not limited to the axial skeleton, with peripheral joint and extra-skeletal symptoms occurring. Extra-skeletal involvement in AS may affect the eye, heart, great vessels, lungs, gastrointestinal tract and genitourinary system.

Uveitis is the most common extra skeletal manifestation and occurs in 20–40% of AS patients. Within the general population, the incidence of ankylosing spondylitis is 0.9%, with a male predominance between 2–3:1. Typical findings of AS include low back pain and stiffness, limited motion in the lumbar spine, limited chest expansion and radiographic findings of sacroiliitis. Morning stiffness is the most sensitive indicator of AS, followed by an onset less than 35 years of age, and chronic symptomatology with over three months of low back pain. Although, the age of onset of symptoms is usually less than 35 years of age, the disease process itself may actually begin as young as eight years of age. Of 29 separate history, physical exam and blood analysis tests, getting out of bed at night and reduced lateral mobility appeared to be the only moderately accurate items in diagnosing AS. The etiology of AS is not well defined, but genetic factors are better predictors of radiographic progression and disability than environmental factors in the disease. The major histocompatibility complex genes, including the human leukocyte antigen...
B27 (HLA-B27), may account for half of the genetic susceptibility in AS.  

Like the etiology, the natural history for the individual patient is poorly defined, with exacerbations and remissions varying from patient to patient.  

AS is a linearly progressive disease, with less than 1% of patients entering long term remission. The prognosis for individuals with active AS over two years is poor and there is no cure for this disease at present. The goal of treatment in AS is to improve the quality of life through pain control and to maintain or improve the patient’s functional abilities. Symptom control has focused on the use of medication and therapeutic exercise. Medication toxicity is common in patients with AS and physiotherapy, including exercise, stretching and modalities have offered inconsistent benefits. 

Even with the present treatments available some patients continue to have severe and inadequately controlled disease. These findings suggest the need for the development of new and more effective treatment options for patients with AS.

Until recently, all reports of ankylosing spondylitis improvement with chiropractic manipulative therapy have been anecdotal. There is limited outcome measure support for the use of chiropractic manipulative therapy in the clinical management of AS. The Bath Ankylosing Spondylitis Functional Index (BASFI) was utilized in a patient with AS undergoing chiropractic treatment and rehabilitation techniques. This was shown to improve this patient’s ability to cope with everyday life. Additionally, chiropractic care resulted in an objective improvement in the SF-36 health survey in a patient with AS, although this survey is not specific for AS. Our case study uses two ankylosing spondylitis specific indices to objectively support the use of chiropractic manipulative therapy in the management of AS. The Ankylosing Spondylitis Disease Activity Index (BASDAI) and the Bath Ankylosing Spondylitis Functional Index (BASFI) are used to document improvement in disease activity and patient function during chiropractic therapy in a patient with AS.

Case report

History

A 34-year-old male student presented with upper back pain and stiffness and low back pain from a long standing history of ankylosing spondylitis (AS). The diagnosis of AS was initially made by an orthopaedic surgeon ten years earlier following six years of bilateral sacroiliac pain and morning stiffness. The diagnosis was established following radiographic examination and blood testing confirmation of HLA-B27 histocompatibility complex. Although, non-steroidal anti-Inflammatory drugs (NSAIDs) had been prescribed to control pain and inflammation, the patient had discontinued NSAIDs because of associated intestinal discomfort three months prior to seeking treatment at our office. After discontinuing prescription NSAIDs, he began taking 0–2 enteric coated acetylsalicylic acid tablets (500 mg) in the evening and glucosamine sulfate with devil’s claw (500mg), three times a day. He was on a regimented exercise program at the time, consisting of daily morning stretches in the shower, gym ball stretches, weight training (three times a week) and aerobic exercise (one hour run, 3–5 times a week). This patient had received chiropractic treatment in the past, including full spine manipulation, on an irregular basis, and had experienced pain relief from the manipulations.

Physical exam

Chiropractic examination revealed a healthy male in his fourth decade of life. Postural evaluation in the coronal plane failed to demonstrate any clinically significant postural changes. Postural evaluation in the sagittal plane demonstrated mild to moderate anterior head carriage with increased upper thoracic kyphosis. Palpation of the regional musculature demonstrated hypertonic and tender trapezius, rhomboid, intercostal, cervical and thoracic paraspinal musculature bilaterally. Active range of motion of the thoracic spine was full, but generally painful throughout. Chest expansion/excursion measured 1.5 cm. Motion palpation revealed painful intersegmental restrictions in movement in the cervical and thoracic spine. Neurological tests for the upper and lower limbs were unremarkable. Deep tendon reflexes were graded 2+ and muscle strength testing was 5/5 throughout. There were no sensory deficits to light touch or pinprick in the upper or lower limbs. Plantar responses were down-going. Orthopaedic examination was unremarkable, aside from pain on posterior to anterior sacroiliac joint compression.
Symptomatic improvement

Imaging
To rule out any clear contraindications to spinal manipulation, lumbar and thoracic radiographs taken three years prior were reviewed. Radiographs of the pelvis and lumbar spine demonstrated characteristic sacroiliac joint ankylosis with subchondral sclerosis (Figure 1) and ankylosis of the left 12th costotransverse joint (Figure 2). There is a mild loss of superior joint space of the right hip (Figure 2). Squaring of the anterior vertebral bodies of the mid thoracic spine is also present (Figure 3). No syndesmophytes were noted in the lumbar or thoracic spine, implying that the disease has not progressed to the end-stage at the discvertebral junction. Since no recent radiographs of the cervical spine were available, a cervical spine Davis series was performed. Lateral radiographs with flexion-extension studies reveal a post-inflammatory ankylosis of C5–6 with vertebral squaring, disc narrowing and facet ankylosis (Figure 4). There is also an associated hypermobility of C4–5. The atlantodental interspace (ADI) is increased, measuring 3mm in flexion. The initial radiology report suggested a C5–6 block vertebrae. In retrospect with the clinical history, the increased ADI, combined with the hazy articular facet joint margins, vertebral body squaring and disc narrowing at C5–6 the reported block vertebrae is most likely post-inflammatory in nature.

Management
History, examination and imaging findings confirmed the diagnosis of ankylosing spondylitis. A therapeutic plan of management including soft tissue therapy to the cervical and thoracic paraspinal musculature and spinal manipulative therapy to the lower cervical, thoracic and lumbar spine, was initiated. Spinal manipulative therapy to the cervical spine was not utilized at C0–C1, C1–C2 or C5–6 segments due to the increase in the ADI and the C5–6

Figure 1  Tilt-up sacroiliac joint view demonstrates obliteration of the articular margins of the sacroiliac joints suggesting bony ankylosis. Subchondral sclerosis of the sacroiliac joints is also noted.
Ankylosed vertebrae. Anterior and posterior rib mobilizations and manipulations were also performed. Interferential current was used to control pain and to facilitate the manipulations and mobilizations. Thoracic manipulation consisted of diversified posterior-anterior (Carver) compressive procedures and the lower cervical and lumbar spine manipulations consisted of diversified rotary manual procedures. The patient was treated three times per week for 18 weeks.

The patient was assessed on four occasions: 0, 5, 10 and 18 weeks. To assess patient changes in function over the eighteen-week treatment period the Bath Ankylosing Spondylitis Functional Index (BASFI) was employed (Table 1). To measure changes in disease activity in response to therapy the Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) was utilized.

**Results**

Prominent decreases in both the BASFI and the BASDAI scores suggest a notable improvement in function, disease activity and symptomatology in this patient with ankylosing spondylitis. For the BASFI, the greatest im-

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**Figure 2** AP lumbar view demonstrates sclerosis of the right sacroiliac joint. The sacroiliac joint margins are not well visualized, suggesting bony ankylosis. There is a suggestion of bony fusion at the left 12th costotransverse joint. Mild superior joint space loss is also observed in the right hip.

**Figure 3** Lateral thoracic view demonstrates vertebral body squaring of the mid thoracic vertebrae.
Symptomatic improvement in function was realized in the first ten weeks of treatment, with a decrease from 0.52 to 0.08. There was also an increase in chest expansion by 2.5 cm and an increase in forward flexion by 4 cm following the course of chiropractic treatment (Table 1).

**Discussion**

The management of ankylosing spondylitis is complicated by an undefined etiology, by exacerbations and remissions and by the linear progression of the disease. Current management strategies may include pharmaceuticals, exercise and diet.

Pharmaceutical therapies for AS include NSAIDs, anti-rheumatic disease modifying drugs and newly developed therapies targeting tumor necrosis factor alpha. There is no consistent evidence in the literature that NSAIDs have a beneficial effect on skeletal mobility and no drug therapies have been shown to prevent structural damage in spondyloarthritis. Our patient chose to discontinue his prescription NSAID use because of gastrointestinal side effects. Other pharmaceutical therapies available at the time may have offered benefit to this patient, but he expressed an interest in decreasing his reliance on pharmaceuticals to manage his condition.

Exercise is traditionally recommended to preserve flexibility, mobility and the upright posture. One study concluded that regular exercise over a 5-year period prevented the decline in spinal mobility. Moderate and regular exercise may be beneficial for both functional status and disease activity, but it may be the consistency and not the quantity of the exercise that is most important. In a systematic review of physiotherapy interventions in AS by Dagfinrud, three robust randomized control trials utilized exercise as the main treatment intervention. The results of these studies were mixed with some short term benefits for pain, stiffness and spinal mobility reported, but there was insufficient evidence to support or refute the use of physiotherapy interventions for AS. The patient in our study, as outlined previously, was on a self regimented exercise program that remained

![Image of cervical flexion view](Image)

**Figure 4** The cervical flexion view demonstrates an increased ADI. There is a suggestion of vertebral squaring, disc narrowing and facet ankylosis visualized at C5–6.
unchanged prior to treatment and throughout the course of treatment. Any exercise benefits in this study were controlled by the patient’s consistent exercise program prior to and during our treatment.

Although, dietary modifications were not attempted in this case, there are reports of patients who use diet to alleviate rheumatic symptoms. In two specific case studies a vegan diet, and a low starch diet provided improvement in AS symptoms. As no dietary modifications were attempted in our study, diet was considered an unlikely confounding factor.

This case supports the use of chiropractic treatment in AS. The patient stated that the treatment caused an increase in symptoms shortly after treatment, but this was followed by a long term decrease in symptoms over the treatment plan. This subjective finding was supported in his BASFI and BASDAI scores over the 18-week course of treatment. The BASDAI consists of six visual analogue questions relating to the five major symptoms pertaining to AS: fatigue, spinal pain, joint pain/swelling, localized tenderness and morning stiffness. The Spondylitis Disease Activity Index has been shown to be quick, simple, reliable \( (r = 0.93; p < 0.001) \), sensitive to change and evaluates the entire spectrum of AS. It also shows good test-retest-reliability and good internal consistency. The BASFI consists of eight questions on activities relating to functional anatomy and two questions assessing the patient’s ability to cope with their life. The questions are answered on a 10 cm visual analogue scale and a mean of the 10 questions gives the overall BASFI score (0–10).

The reproducibility of the BASFI is good \( (r = 0.89, p < 0.001) \) and has been shown to be sensitive in documenting improvement in the functional ability of patients over a 3-week period (mean score change = \(-1.07, p = .004\)).

Patient’s BASFI scores compared well with functional tests performed by an external observer. For this case study, the BASFI was chosen since it is thought to be more responsive than the Dougados Functional Index (DFI) and the AS specific version of the Health Assessment Questionnaire (HAQ-S). Additionally the BASFI has been shown to be superior in detecting changes in functional performance, is sensitive to change across the whole spectrum of the disease and is quick and easy to complete. Chest expansion and finger-tip-to-floor distance were also assessed to measure the disease severity in AS. In addition to improvement in both the BASFI and BASDAI, improvements were also noted in chest expansion and finger-tip-to-floor distance. These tests are empirical and objective in nature and support the efficacy of chiropractic therapy, including chiropractic manipulative therapy, in increasing function and decreasing disease activity in this patient with AS.

Chiropractic manipulative therapy may be an effective treatment option to decrease pain and increase joint function in patients with AS. However, patients with AS may be discouraged from seeking chiropractic manipulative therapy by their physician or by their support society. A number of risk factors must be considered to ensure safe utilization of chiropractic manipulative therapy in the management of AS. Patients with AS may have

<table>
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<th>Time in Weeks</th>
<th>BASDAI*</th>
<th>BASFI**</th>
<th>Chest Expansion (cm)</th>
<th>Finger to Floor test (cm)</th>
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*Bath Ankylosing Spondylitis Disease Activity Index

**Bath Ankylosing Spondylitis Functional Index
acutely inflamed joints and chiropractic practice guidelines advise that manipulation of acutely inflamed joints is contraindicated.38 Osteoporosis may occur early in the disease, predisposing patients to an increase in vertebral compression fractures and traumatic spinal fractures of the cervical spine.39 There is also an increased risk of traumatic spinal cord compression, which is estimated to be 11 times greater in the AS population compared to the population at large.40 It has been suggested that the prevention of sudden neck movements is of vital importance in patients with ankylosing spondylitis of the cervical spine.41 There is one reported case of paraplegia following chiropractic manipulation of a patient with ankylosing spondylitis.42 While these risk factors are considerable, they are present in only a minority of AS cases. However, in subacute and chronic cases of AS, without local indications of ligamentous laxity, anatomic subluxation or ankylosis, chiropractic manipulation is not contraindicated.38 The authors agree with the cautious use of joint manipulation in AS, however in this case, spinal manipulation of the non-contraindicated joints was continued throughout the 18 weeks of treatment.

It is important to re-assess the cervical spine of patients with AS who are receiving cervical spinal manipulation. AS may affect the cervical spine and with progression may result in a decrease in neck motion and an increase in cervical kyphosis.43 In a study of 61 AS patients in the Moroccan population, 70% had a history of cervical night pain with associated morning stiffness.43 Radiological involvement of the cervical spine was observed in 54%, with vertebral body squaring and facet joint involvement occurring most frequently. The cervical spine radiological and clinical involvement increased with age and disease duration. The prevalence of cervical spine radiographic abnormalities was: 19.6% after 5 years, 29.9% after 10 years, 45.1% after 15 years and 70% after 20 years. Of the 61 Moroccan AS patients observed, 12 patients with neck pain presented with no cervical radiological signs and notably, two patients with cervical radiological signs did not report neck pain. Since neck pain does not adequately predict radiographic changes, there is a need for frequent reassessment of the cervical spine of patients with AS to rule out local contraindications to cervical spinal manipulative therapy.

Spontaneous atlantoaxial subluxation (AAS) may be a serious complication in rheumatic disorders including seronegative spondyloarthropathies like AS.44,45,46 This poses a considerable risk for upper cervical manipulation. The transverse ligament instability is a potentially life threatening complication that has traditionally been considered a late finding in AS.45 However, current studies contradict the notion that AAS is a late finding in the disease.44,47,48 and has been an early presenting complication in some patients with AS.47,48 Furthermore, the presence of atlantoaxial subluxation is not associated with an increase in the duration of the disease.44 The prevalence of AAS was investigated in 103 consecutive patients with a 10-year mean AS duration and there was a 21% anterior AAS and a 2% vertical AAS prevalence.44 This finding is higher than previously reported in other studies, which have reported a 2–15% atlantoaxial incidence.49,50,51 Anterior AAS was shown to be associated with an increase in the degree of radiological sacroiliitis, while other factors, including disease duration, peripheral arthritis, current symptoms, functional index, use of steroids and HLA-B27 were not statistically associated with anterior AAS.44 Thus, clinical symptoms do not appear to predict the presence of AAS, and only radiographic findings are shown to be robust. A two-year followup of the patients with AAS by Ramos-Remus52 revealed that 32% of patients with AAS showed radiological progression with or without neurological symptoms.

AAS may produce a number of different neurological signs and symptoms, but most AS patients with AAS present with little or no neurological signs and symptoms at all.44 The lack of signs and symptoms of AAS may mask the underlying pathology, creating a potential for disaster if managed inappropriately by the chiropractor. Ligamentous instability, such as AAS, is a local absolute contraindication to high velocity thrust procedures.38 Presently, there is no agreement in the literature on the minimal distance required to diagnose anterior AAS. AAS has been suggested if the ADI is 4 mm or greater.44 Recently, AAS was diagnosed clinically with an ADI equal to or greater than 3 mm.43 Our patient who has confirmed AS, a probable cervical spine involvement at C5–C6, and an ADI of 3 mm suggests AAS. Hence, manipulation of C0–C1, C1–C2 and C5–C6 spinal segments were considered a contraindication in this case. The patient in our study had received upper cervical manipulations within the year prior to his presentation to the authors. Lateral cervical spine radiographs obtained in
flexion are essential to bring this clinical issue to the foreground and decrease the patient’s risk associated with upper cervical spine manipulation. The increased incidence and the potentially silent nature of the transverse ligament involvement, with few clinically significant neurological symptoms, combined with our clinical experience, highlight the importance of obtaining lateral cervical spine flexion radiographs in patients with AS. The authors of this case study advise that flexion radiographs be considered for all patients with AS, prior to upper cervical spine manipulation, regardless of disease duration or current symptoms. The increase in ADI creates a local contraindication to manipulation, but as suggested in this case, patients with local contraindications to manipulation may still benefit from manipulation of other joints. Thus, these patients should not be discouraged from seeking chiropractic manipulative therapy.

This study was limited in sample size, experimental design and by uncontrolled variables. Specifically other modalities such as soft tissue therapy, rib mobilizations and interferential current were used in addition to chiropractic spinal manipulation and mobilization. The soft tissue therapy and interferential current were reported by the patient to decrease the initial sensitivity associated with receiving spinal manipulation. An attempt was made to ensure that this patient’s exercise program, diet and supplement intake remained constant throughout, but some variability may have occurred over the 18 weeks of study. As well, ankylosing spondylitis is characterized by periods of exacerbations, which may further confound the results. Specifically, it is not possible to prove that the change in BASDAI or BASFI was a treatment effect and not simply an improvement or remission in symptoms following an exacerbation. The patient in our study was functioning at a high level prior to beginning treatment, as indicated by a low baseline BASFI score. This created a situation where there was less range to measure the treatment effect. So unlike the BASDAI, the BASFI values were limited to the lower end of the scale, ranging from 0.52 to 0.08, pre and post treatment. However, an important goal of treatment is to maintain and/or improve function in patients with AS, and even a small change in the BASFI score is a favorable outcome. As well, functional disability is the most important patient outcome in decreasing the financial costs associated with AS. In retrospect, continued follow-up assessments possibly at 6 and 12 months would have been helpful to determine if these benefits were long lasting. However, the results of this case study suggest that chiropractic manipulation and mobilization may improve function, disease activity and symptomatology in patients with AS.

Chiropractic manipulative therapy has been well supported as a useful adjunct to limit pain and improve function in individuals with non-specific back pain. In this patient, chiropractic manipulative therapy has also been shown to be a useful adjunct for the treatment of specific back pain and symptomatology due to ankylosing spondylitis. Chiropractic therapy has facilitated an increase in function and a decrease in disease activity in this patient with AS. Therefore, chiropractic manipulative therapy may provide an integral and effective treatment option in the clinical management of this disease. Further research is required to substantiate a conclusive cause/effect relationship between chiropractic manipulative therapy and improvement in individuals with ankylosing spondylitis.

Conclusion

An improvement in the Bath Ankylosing Spondylitis Functional Index and the Bath Ankylosing Spondylitis Disease Activity Index in a patient with long standing AS was a clinically favorable and noteworthy outcome. Specifically, these indices provide unprecedented support for the effectiveness of chiropractic manipulative therapy in the treatment of AS in this case. Validated instruments for function and disease activity improve the assessment of new therapeutic treatment options in AS, which may ultimately improve the treatment efficacy and patient outcomes in the clinical management of AS. More intensive research utilizing these instruments on patients with AS undergoing chiropractic therapy is suggested.

References

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