Multimodal Chiropractic Care of Pain and Disability for a Patient Diagnosed With Benign Joint Hypermobility Syndrome: A Case Report

Richard G. Strunk DC, MS^{a,*}, Mark T. Pfefer RN, DC, MS^b, Derrick Dube^c

^a Assistant Professor, Research Clinician Cleveland Chiropractic College, Overland Park, KS

^b Professor, Research Director Cleveland Chiropractic College, Research, Overland Park, KS

^c Student, Cleveland Chiropractic College, Overland Park, KS

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Abstract

Objective: The purpose of this case report is to describe multimodal chiropractic care of a female patient diagnosed with benign joint hypermobility syndrome (BJHS) and a history of chronic spine pain.

Clinical features: A 23-year-old white female presented for chiropractic care with chronic low back pain, neck pain, and headaches. The patient was diagnosed with BJHS, including joint hypermobility of her thumbs, elbows, right knee, and lumbopelvic region. A 6-year history of low back pain and varicose veins in her posterior thighs and knees were additional significant diagnostic findings of BJHS.

Interventions and outcomes: The treatment consisted of spinal and extremity manipulation, Graston technique, and postisometric relaxation combined with sensory motor stimulation and scapular stabilization exercises. The patient was seen 15 times over an 18-week period. After 18 weeks of care, the Revised Oswestry Low Back Questionnaire and Headache Disability Index demonstrated clinically important improvements with her low back pain and headache; but little change was noted in her neck pain as measured by the Neck Disability Index.

Conclusion: This patient with BJHS who had decreased disability and spine pain improved after a course of multimodal chiropractic care.

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Introduction

Benign joint hypermobility syndrome (BJHS) is a hereditary connective tissue disorder defined by pain and hypermobility in multiple joints.¹⁻⁴ Benign joint hypermobility syndrome is also known as *joint*

* Corresponding author. MS-10850 Lowell Ave, Overland Park, KS 66210. Tel.: +1 913 234 0798; fax: +913 234 0926.

E-mail address: Richard.strunk@cleveland.edu (R. G. Strunk).

hypermobility syndrome and *hypermobility syndrome*. Furthermore, it is important to be aware that BJHS shares similar clinical features to Ehlers-Danlos syndrome–hypermobility type(EDS-HT). Both BJHS and EDS-HT can present with generalized joint hypermobility, abnormal skin features, recurring joint dislocations, chronic joint/extremity pain, and a positive family history. The close association between BJHS and EDS-HT causes many to view them as the same condition.^{5,6} However, with the lack of readily available clinical testing, the association between BJHS and EDS-HT is not definitive. Benign joint hypermobility syndrome is considered to be more common in African, Asian and Middle Eastern populations and is also more common in females and younger individuals.^{4,7-11} The true prevalence of BJHS is unknown, but estimates range from 5% in the United States to 43% in African populations.^{7,12} It is diagnosed by patient history and/or physical examination findings such as generalized joint hypermobility, chronic limb and or back pain, and connective tissue lesions. These history and examination findings are grouped into “major” and “minor” criteria called the *Brighton criteria* (Fig 1).¹³ Within the Brighton criteria, the amount of joint hypermobility is calculated through the Beighton score (Fig 2). The Beighton score is used to quantify the extensiveness of

Major Criteria

- Beighton score of ≥ 4 (Fig. 2)
- Arthralgia for longer than 3 months in 4 or more joints

Minor Criteria

- Beighton score of 1, 2, or 3 (Fig. 2)
- Arthralgia (>3-month duration) in 1-3 joints or back pain (>3-month duration) or spondylosis, spndylolysis/spondylolisthesis.
- Dislocation or subluxation in more than one joint, or in one joint on more than one occasion.
- Three or more soft tissue lesions (eg, epicondylitis, tenosynovitis, bursitis)
- Marfanoid habitus (tall, slim, span greater than height [>1.03 ratio], upper segment less than lower segment (<0.89 ratio), arachnodactyly)
- Skin striae, hyperextensibility, thin skin, or abnormal scarring
- Ocular signs: drooping eyelids, myopia, antimongoloid slant
- Varicose veins, hernia, or uterine or rectal prolapse, mitral valve prolapse

Requirement for diagnosis

BJHS is diagnosed in the presence of 2 major criteria, one major criterion plus 2 minor criteria, or 4 minor criteria. Two minor criteria will suffice where there is an unequivocally affected first-degree relative in family history. The syndrome is excluded by the presence of Marfan or Ehlers-Danlos syndrome (other than the hypermobility type of Ehlers-Danlos syndrome, formerly EDS III).

Fig 1. Brighton criteria for the diagnosis of BJHS.

Passive dorsiflexion of the fifth metacarpophalangeal joint to at least 90°.

(1 point for left; 1 point for right)

Passive opposition of the thumb to the volar aspect of the ipsilateral forearm

(1 point for left; 1 point for right)

Active hyperextension of the elbow to at least 10°

(1 point for left; 1 point for right)

Active hyperextension of the knee to at least 10°

(1 point for left; 1 point for right)

Lumbopelvic forward flexion with placement of hands flat on the floor without bending the knees (1 point)

One point is gained for each side of the body for the first four maneuvers listed above, such that the hypermobility score is a maximum of 9 if all are positive.

Fig 2. Nine-point Beighton score for joint hypermobility.

joint hypermobility on a “0 to 9” scale where 0 indicates no hypermobility and 9 indicates joint hypermobility in multiple body sites such as the low back, elbows, hands, and knees. Studies have shown adults and children with BJHS frequently experience arthralgia, with adults also being prone to generalized back pain.¹⁴⁻²¹ There are also some studies that suggest BJHS is associated with fibromyalgia,^{22,23} and a prospective cohort study concluded that joint hypermobility in adolescence was a risk factor for musculoskeletal pain at the shoulder, knee, and ankle/foot joints.²⁴

Research on the conservative treatment of BJHS is quite limited, with most high-level studies (randomized controlled trials) focused on knee pain and low-level studies (single-group prospective cohort, case series, case reports) focused on the spine or full body pain.²⁵⁻³³ Proprioceptive or balance exercises have been the most widely studied treatment approach, showing some promise for knee pain.²⁵⁻²⁷ As a result of the limited amount of high-level studies, standardized treatment guidelines are not available, with treatment recommendations based on low levels of evidence.³⁴ It is generally recommended that, for patients with BJHS and chronic pain, a multidisciplinary approach consisting of physiotherapy (strengthening and proprioceptive exercises), cognitive behavioral therapy, and pharmacology is best.

Regarding research on chiropractic spinal manipulative therapy and patients with spinal pain and BJHS, only 3 case reports have been published.^{28,32,33} Despite the limited amount of research, the results from these case reports showed a pain reduction from multimodal chiropractic care. The small quantity of clinical research on the conservative management of BJHS and pain calls for more research on this topic. The purpose of this case report is to describe multimodal

chiropractic treatment of a patient diagnosed with BJHS and spinal pain.

Case Report

Patient History

A 23-year-old white female presented with persistent headaches and right neck and low back pain. All areas were rated a 4/10 for average pain, 5/10 for worst pain, 1/10 for best pain, and 2/10 for pain at time of initial visit using the numeric pain rating scale. The numeric pain rating scale is commonly used and has shown to be reliable and valid.^{35,36} Her neck pain was achy and sometimes sharp, localized to the right upper trapezius with referral into the right upper neck and shoulder region. The neck pain began insidiously 9 months ago while working on patients at a dental hygienist school. Resting helps decrease the pain, whereas seated postures at the dental school increase the pain. The Neck Disability Index (NDI) was scored at 10% at the initial visit. The NDI is a 10-item questionnaire with good reliability, as well as good construct and concurrent validity in an ambulatory clinic population.³⁷ It has also been shown to possess stable psychometric properties and is thought to provide an objective means of assessing the disability of patients with neck pain.³⁸

Her dull achy headaches were localized to the suboccipital and temporal area and occurred with the neck pain at least 3 times per week. No photophobia/phonophobia or other symptoms were reported with the headaches. The 25-item Headache Disability Index (HDI) score was 12%. The HDI questionnaire is designed to measure the disability levels among patients with headache. It has been shown to have strong internal reliability and construct validity as well as acceptable levels of test-retest reliability.³⁹ The deep and dull low back pain was localized to the right lower lumbar spine and sacroiliac joint. The pain that began 6 years ago from childbirth is made worse by prolonged standing and sitting and temporarily decreased by hot showers and previous chiropractic care. The patient's initial Revised Oswestry Low Back Questionnaire (OLBQ), which measures low back pain disability, was 16%. This 10-item questionnaire has demonstrated good reliability⁴⁰ and has shown to be responsive to clinically meaningful change.⁴¹⁻⁴⁵ Except for a horse-back riding fall as a child, a low weekly exercise level and a low daily fruit and vegetable intake, the patient's past medical history and health habits were unremarkable.

Initial Examination

The orthopedic examination of the cervical spine and lumbopelvic regions indicated mechanically induced pain in both regions. In the cervical spine, pain was reproduced with stretching tests (shoulder depression/costoclavicular test) in the right upper trapezius muscle. Lumbopelvic pain was reproduced at the lumbosacral junction and right sacroiliac joint with lumbosacral extension tests. Cervical and lumbar active and passive range of motion demonstrated no limitations in motion or pain except for sharp and deep pain at the right sacroiliac joint and lumbosacral junction with active lumbar extension. Postural examination revealed a forward flexion bias as observed by anterior head carriage, internally rolled shoulders, anteriorly translated torso and anteriorly rotated pelvis in the sagittal plane. The Janda muscle movement pattern testing⁴⁶ demonstrated bilateral hyperactivity of the upper trapezius muscles, whereas the motion palpation endplay examination⁴⁷ revealed joint hypomobilities in the cervical, thoracic, and lower lumbar spinal joints as well as the right sacroiliac joint. The soft tissue palpation examination indicated multiple areas of hypertonicity and tenderness in the thoracic and lumbar paraspinals, as well as the right suboccipital, bilateral levator scapula, bilateral sternocleidomastoid, and right upper trapezius muscles. The patient was initially diagnosed with cervical/lumbar myalgia; cervicogenic headaches; sacroiliac arthralgia; and joint dysfunction of the cervical, thoracic, and lumbopelvic regions.

Examination for BJHS

Because BJHS examination procedures were not part of the standard examination performed on the initial visit by interns, a screening examination (Beighton score and Brighton criteria) for BJHS was performed on the patient's third visit because of the patient's age, sex, and history of spinal pain. The screening examination was later confirmed by a focused BJHS examination with goniometry on the patient's seventh visit. The patient's Beighton score was a "6" out of "9." Her elbows, knees, and fifth metacarpophalangeal (MCP) joint hypermobility was measured with a 2-arm goniometer, whereas thumb opposition and lumbopelvic flexion were visually observed. Left and right elbow (humeroulnar joint) hyperextension was 20° (Figs 3 and 4), right knee hyperextension was 10° (Fig 5), left knee hyperextension was 9°, left fifth MCP dorsiflexion was 85°, and

Fig 3. Left elbow extension. (Color version of figure is available online.)

Fig 5. Right knee extension. (Color version of figure is available online.)

right fifth MCP dorsiflexion was 75°. Visually, both the left and right thumbs displayed full passive opposition to each respective forearm (Figs 6 and 7), and lumbopelvic flexion was full with palms flat on the floor and knees fully extended (Fig 8). The physical examination of the patient's legs revealed varicose veins bilaterally in the posterior knee region and distal quarter of the posterior thigh.

The diagnosis of BJHS was made by 1 major Brighton criterion (Beighton score = 6) and 2 minor Brighton criteria (varicose veins and a 6-year history of low back pain).

Treatment

Multimodal treatment consisted of joint manipulation and soft tissue/muscle energy treatment combined with scapular and sensory motor stimulation exercises. The most common type of manipulation (visits 4 through 15) administered was a mechanical-force, manually assisted technique applied by the Activator IV adjusting instrument to the spine, pelvis, and extremities.⁴⁸ This

instrument allows for a specific, low-force impulse to the vertebral joint and may be preferred in patients with connective tissue fragility and systemic joint hypermobility.^{28,49} High-velocity, low-amplitude spinal manipulation to the cervical, thoracic, and lumbar spine (HVLA SM) and drop technique to the pelvis⁴⁷ were performed on the second and third visits before the patient was diagnosed with BJHS. Because of the patient's new diagnosis of BJHS on the third visit and minor symptomatic change from treatment, the Activator method was used subsequently. In conjunction with the spinal manipulative techniques (Activator method and HVLA SM), a soft tissue technique was used each

Fig 4. Right elbow extension. (Color version of figure is available online.)

Fig 6. Right thumb hypermobility. (Color version of figure is available online.)

Fig 7. Left thumb hypermobility. (Color version of figure is available online.)

visit to the upper trapezius and upper thoracic paraspinal muscles. Graston technique was used for the first 8 visits, and then postisometric relaxation technique was applied at the remaining visits to help reduce hypertonicity.⁵⁰ Reevaluation of patient progress was performed 3 times throughout care; and because of the results of functional tests at these reevaluations, active care exercises were administered. Because of a failed “Vele’s Reflex Stability Test of the Transverse Arch” and the inherent hypermobility/laxity of the knees and low back, approximately 8 minutes of sensory motor training using a wobble board was used each visit

Fig 8. Trunk flexion hypermobility with palms flat on the floor and knees fully extended. (Color version of figure is available online.)

starting on the ninth visit. The sensory motor training was initially performed with the goal of maintaining a neutral posture without any external stimulation. Once the 2-legged neutral posture was maintained without much difficulty, the patient was progressed to playing catch using medicine balls while maintaining an upright posture on the wobble board. The degree of difficulty with the medicine ball catch game was increased over the last 6 to 7 weeks by using a graded approach starting with 2-lb balls and concluding with 6.6-lb balls.

At about the same time as the sensory motor training was introduced, the patient was given home balance exercises and scapular stability exercises. The home balance exercises were to be done using a beginner-level foam stability pad. The patient was instructed to hold a neutral posture while performing a single-leg stance on the stability pad for 5 to 10 min/d. Once the single-leg stance exercise became easy, the patient was instructed to progress to performing hip extension, flexion, and abduction exercises while maintaining a neutral upright posture for 5 to 10 min/d on the same stability pad. To facilitate stability of the scapula, seated scapula depression/retraction exercises and serratus anterior wall push-up plus facilitation exercises were given to the patient to perform on a daily basis. For both scapular exercises, the patient was instructed to perform 1 set of 15 to 20 repetitions and progress to more and in the prone position (for retraction/depression) as the difficulty decreased. Other recommendations made to the patient were to increase weekly aerobic exercise levels to 4 times a week and increase daily fruit and vegetable serving intake to 3 times a day. All examinations (except for the BJHS examinations) and treatment were performed at a chiropractic college outpatient clinic by qualified chiropractic interns under the direction of a licensed doctor of chiropractic. The BJHS examinations were performed by the lead author.

Outcomes

The HDI, NDI, and the Revised OLBQ were administered at follow-up intervals of 8, 15 and 18 weeks. The patient received treatment approximately over an 18-week period where she averaged 1 visit per week for a total of 15 visits despite missing multiple visits during the first couple of months of care including a 3½-week period of no care during the second month. The HDI and Revised OLBQ scores improved the most from the start of care to week 18 (Table 1). Whereas the Revised OLBQ demonstrated a clinically meaningful improvement (a decreased of 12 points),^{41,45} the NDI increased at week 8 and

Table 1 Patient Outcomes at Baseline and Follow-Up Intervals

	Baseline	8 wk	15 wk	18 wk
HDI (%)	12	8	2	0
NDI (%)	10	18	8	8
Revised OLBQ (%)	16	10	8	4

Revised OLBQ, HDI, and NDI: 0% to 100%, with 0% = no disability and 100% = severe disability.

HDI, head disability index; NDI, neck disability index; OLBQ, Oswestry Low Back Questionnaire; wk, weeks.

demonstrated little change at weeks 15 and 18. The Patient Global Impression of Change scale (PGIC) was administered at weeks 15 and 18. At week 15, the scale indicated that the patient was “much better”; and at week 18, it indicated that the patient was “a little better.” The PGIC is a self-reported 7-point Likert scale where a patient assesses his or her degree of change since starting treatment, ranging from very much better to very much worse. The PGIC has been well validated and has been commonly used by pain researchers as a standard outcome instrument.⁵¹⁻⁵⁴ Although the patient reported exercising 4 times a week and lessened her daily “carbohydrate/sugar” levels, it is not known how compliant the patient was with the home balance and scapular stability exercise program. The patient gave consent for this study to be published.

Discussion

This is one of the first reported cases of the chiropractic management of a patient with BJHS. The results of the outcome indices showed that the patient had a clinically important improvement in her headache and low back disability levels over an 18-week course of multimodal chiropractic care. Although the patient’s headache and low back pain improved, the patient’s neck disability levels varied since the start of care. The varied outcome seen with this patient has also been observed in other patients who have this condition as documented in a retrospective single-group cohort study³¹ and in other case reports.^{28,29,32,33}

Patients with BJHS typically display a fluctuating pain pattern characterized by exacerbations and remissions and many times do not make a full recovery from treatment. Even with the varied outcome, this case report is important to health care providers especially doctors of chiropractic because of the low number of case reports and nonexistence of experimental research

on effective conservative management of this condition (related to the spine) or similar conditions.^{28,29,32,33}

Diagnosis of BJHS

Because of the reported high prevalence rates of BJHS in certain populations and the underdiagnosis of BJHS,⁶¹ screening for BJHS would likely add value in determining the best course of treatment for patients with chronic pain and this entity. Hakim and Grahame⁶² developed a simple 5-item questionnaire to screen patients for joint hypermobility. The questionnaire has been shown to have good sensitivity and specificity for hypermobility in patients when 2 or more of the responses to the questions are affirmative. When screening with this questionnaire, a health care provider would not have to use the Brighton criteria on every patient with pain, just on patients with 2 or more affirmative responses on the questionnaire. Screening for BJHS might be helpful for patients with chronic pain who do not respond to an initial module of care. Practitioners who practice spinal manipulative procedures might apply specific techniques to this population of patients with better results if they know their patient has BJHS. Because of the nature of BJHS where articular hypermobility is present and joint stability is uncertain, HVLA SM represents a relative contraindication to the involved area of pathology.⁴⁹ More research is needed to determine the best course of treatment for patients with BJHS.

Limitations

Without intervention, headaches, neck pain, and low back pain tend to follow a recurrent and persistent natural course.⁵⁵⁻⁶⁰ It is possible that the patient may have improved because of the natural course of the disorder or other factors that were not measured or that we are unaware of. Because this was a single case report, it is not appropriate to generalize the effects from this patient to other patients with BJHS and spinal pain. Further observation and experimental research with larger sample sizes are needed to determine what the effects multimodal chiropractic care has on patients with BJHS and chronic spinal pain.

Conclusion

This case report describes the response to multimodal chiropractic treatment of a patient diagnosed with BJHS

and low back pain, neck pain, and headaches. The patient experienced clinically important improvements in her low back and headache disability levels posttreatment.

Funding Sources and Conflicts of Interest

No funding sources or conflicts of interest were reported for this study.

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