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Chiropractic Care of an 8-Year-Old Girl With Nonorganic, Primary Nocturnal Enuresis: A Case Report

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Abstract

Objective: The purpose of this case report is to describe the chiropractic management of an 8-year-old girl with nonorganic, primary nocturnal enuresis.

Clinical Features: An 8-year-old female patient presented to a chiropractic clinic with persistent nighttime bedwetting. The patient experienced enuresis, on average, 7 nights per week. The patient presented with no other comorbidities or complaints, such as low back or pelvic pain.

Intervention and Outcomes: Chiropractic treatment included high-velocity, low-amplitude manipulation of the left sacroiliac joint over 3 visits. Follow-up at 3 months revealed only 3 subsequent episodes of nocturnal enuresis.

Conclusion: This patient reported the resolution of nonorganic, primary nocturnal enuresis after receiving a series of side-posture chiropractic manipulations of the left sacroiliac joint.

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Introduction

Nocturnal enuresis is involuntary voiding of urine during sleep, in the absence of organic disease, in individuals at a developmental age of 5 years or older.¹ It is a relatively common complaint, with an estimated prevalence of around 15% to 20% of 5-year-olds, 5% of 10-year-olds, and 1% of those 15 years and older.¹

Nighttime bedwetting is a socially disruptive and emotionally stressful condition that may bring stigma, stress, and inconvenience to both those with nocturnal enuresis and their families.¹

The etiology of nocturnal enuresis is unclear.¹ Suggested possible causes of enuresis include a diverse range of factors (eg, physiological, psychological, genetic, neurologic developmental delay, and consumption of foods or drinks with diuretic effects). Unsurprisingly, there is a correspondingly diverse range of conventional and complementary interventions available to individuals with nocturnal enuresis.

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Conventional interventions include pharmacological interventions (eg, desmopressin, tricyclic and related drugs, and other drugs),²⁻⁴ and simple and complex behavioral interventions (eg, reward systems, fluid deprivation, and alarms).⁵⁻⁷ Less traditional interventions include acupuncture, chiropractic, dietary and restricted foods regimens, homeopathy, and hypnosis.¹ It is also important to note that about 15% of children with enuresis become dry each year without any treatment whatsoever.⁸

Although there is limited anecdotal evidence suggesting that children with enuresis may benefit from chiropractic care,⁹⁻¹² systematic reviews of randomized controlled trials (RCTs) have concluded that there is insufficient or inconclusive evidence for the effectiveness of chiropractic intervention for nocturnal enuresis.^{1,13-15} These systematic reviews are based on the 2 RCTs that have been published to date.^{16,17} Unfortunately, both of these trials suffer from methodological shortcomings that substantially limit the conclusions that can be drawn from them.

Inconclusive empirical evidence and positive anecdotes warrant further investigations of chiropractic care for nocturnal enuresis. Therefore, the purpose of this case report is to describe the chiropractic management of an 8-year-old girl with nonorganic nocturnal enuresis. We also provide a review of the current best evidence of chiropractic intervention for nocturnal enuresis and recommendations for future clinical trials.

Case Report

An 8-year-old, white female patient presented with her mother to a chiropractic clinic seeking a second opinion for persistent nighttime bedwetting. The patient reported not having experienced a dry night and denied any history of trauma, accidents, or major falls or injuries. The patient presented with no other comorbidities or complaints such as low back or pelvic pain.

The patient had received standard medical care from her general practitioner, as well as from specialists at the pediatric department at the local hospital. Urine tests and magnetic resonance imaging of the lumbar spine and pelvis had not revealed any abnormalities, and organic or pathological cause had been identified. The patient had been treated with conventional pharmacotherapy (60 µg desmopressin [Ferring Pharmaceuticals, Germany] once per day for 3 weeks followed by 60 µg desmopressin twice per day for 1 week) without any change in bedwetting. In consulta-

tion with her general practitioner, the patient had discontinued medication because of adverse reactions (stomach pain). Behavioral interventions such as fluid deprivation, scheduled waking, and alarms had been trialed for several weeks without any change in nocturnal enuresis.

The chiropractic consultation included an unremarkable neurological and orthopedic examination. Joint motion palpation of the spine was similarly unremarkable, except for a moderate restriction of posterior to anterior glide of the left sacroiliac joint (SIJ). There was also marked tenderness to palpation over the left SIJ.

Chiropractic treatment consisted of gentle isometric pressure over the left paraspinal soft tissues at the lumbosacral junction, followed by a single high-velocity, low-amplitude (HVLA) manipulation of the left SIJ. The patient was positioned on her right side in a basic lumbar roll position, and the SIJ manipulation involved a soft pisiform contact immediately inferior to the patient's left sacral ala with a force vector in the posterior to anterior direction. No additional interventions or specific take-home instructions were provided at this stage.

Immediate reexamination of the left SIJ revealed increased joint motion and diminished tenderness to palpation. During a follow-up consultation 1 week later, the patient reported no bedwetting since the first consultation. On examination, the SIJs exhibited normal joint motion, and no further treatment was provided.

Nine days later, however, the nocturnal enuresis relapsed, and the patient experienced 8 subsequent wet nights before again presenting to the chiropractic clinic. The patient denied experiencing any falls or injuries before the relapse. Similar to the initial consultation, the chiropractic examination revealed a restriction of posterior to anterior glide of the left SIJ accompanied by tenderness to palpation over the left SIJ. The patient received a single HVLA manipulation of the left SIJ on 2 occasions, 8 days apart, and the bedwetting ceased once again after the second treatment. Thus, from the initial presentation to the final discharge 3 weeks later, the patient received a total of 3 HVLA manipulations of the left SIJ.

Follow-up phone calls to the patient's mother at 1 and 3 months after the last visit revealed only 4 subsequent episodes of nocturnal enuresis. The patient's mother reported that the patient's behavior had changed substantially over the last 3 months, including engaging in more play and sleeping over with friends without fear of being wet. The patient's mother consented to have her daughter's personal health information published without divulging personal identifiers.

Discussion

The present case report describes the resolution of persistent, nonorganic nighttime bedwetting in an 8-year-old female patient following chiropractic care consisting of HVLA manipulation of the patient's left SIJ.

Biological Plausibility

Insofar nocturnal enuresis is related to mechanical dysfunction of the spine, there may be a role for chiropractic spinal manipulative therapy. The objective of this section is not to provide an exhaustive review of the relevant neuroanatomy and physiology, but to illustrate the importance of using a plausible biological mechanism and working hypothesis when conducting clinical investigations into the purported effectiveness of spinal manipulative therapy for nonorganic nocturnal enuresis.

The urinary bladder receives both autonomic and somatic nerve supply emerging from the lumbopelvic region of the spine.¹⁸ The parasympathetic control of the bladder (ie, muscle contraction which causes bladder emptying) originates with preganglionic neurons in the sacral spinal cord segments (S2-S4). These fibers travel in pelvic splanchnic nerves and innervate postganglionic neurons in parasympathetic ganglia in or near the bladder wall. The sympathetic control of the bladder (ie, muscle contraction of the internal urethral sphincter to prevent bladder emptying) originates in the lower thoracic and upper lumbar spinal cord segments (T10-L2). The preganglionic fibers synapse in the inferior mesenteric ganglion and the ganglia of the pelvic plexus, and the postganglionic neurons continue in the hypogastric and pelvic nerves to the bladder. The somatic (voluntary) control of the external urethral sphincter (ie, tonic muscle contraction to prevent bladder emptying) originates in ventral horn cells in the sacral spinal cord segments (S2-S4). In addition to the motor supply, there are 3 sets of afferent nerves supplying sensory information from the bladder and urethra,¹⁹ the most important of which, with respect to micturition, are A-delta and C fibers, which convey signals from mechanoreceptors and nociceptors in the bladder wall.

There is evidence that primary nocturnal enuresis is caused by a delay in the maturation of normal bladder function²⁰ and, furthermore, that strong genetic factors cause or predispose children to this developmental delay.²¹ This developmental delay appears to be associated with increased parasympathetic activation of the urinary bladder.^{22,23} It is unknown, however, whether biomechanical dysfunction of the spine or SIJ

can alter the activity of the autonomic and somatic neurons supplying to the urinary bladder and urethra.

The mechanisms responsible for the therapeutic effects of spinal manipulative therapy remain unclear. However, emerging neurophysiological investigations suggest that spinal manipulative therapy may produce a sustained change in the synaptic efficacy of central neurons by activating several types of mechanosensitive paraspinal sensory neurons.^{24,25} As such, there may be differences between manipulation of the lumbopelvic spine vs other regions of the spine in the treatment of enuresis.

Critical Appraisal of the Current Best Clinical Evidence

Systematic reviews of RCTs conclude that there is insufficient or inconclusive evidence for the effectiveness of chiropractic intervention for nocturnal enuresis.^{1,13-15} This conclusion is justified because only 2 clinical trials have been published to date^{16,17} and, moreover, both suffer from methodological shortcomings. What follows is a critical appraisal of these 2 trials, with a focus on assessing the risk of bias. This appraisal will in turn lead to recommendations for future RCTs of chiropractic intervention for nocturnal enuresis.

First, although both RCTs appear to have recruited an adequately representative sample of the target population, they have provided no clear description of either the method used to generate the random allocation sequence or the method used to conceal it in advance of (or during) enrolment of participants. However, incomplete or inadequate reporting does not entail that the design and conduct of these trials were actually deficient. Although true randomization typically generates relatively equal group sizes, both trials ended up including more participants in their active intervention groups (58%¹⁶ and 63%¹⁷). Further indication of possible subversion of the allocation procedure is suggested by the fact that there were significant baseline imbalances between intervention and control groups in both RCTs (eg, intervention and control groups were not comparable at baseline in regard to severity of bedwetting). Thus, although the description of the generation and implementation of the random allocation sequence is unclear, there is sufficient reason to believe that selection bias is present in both trials.

Second, study participants, intervention providers, and outcome assessors appear to have been unblinded in both RCTs. It is generally accepted that it is often difficult or impractical, or in some cases even

impossible, to achieve complete blinding in trials assessing nonpharmacological interventions.²⁶ Although lack of blinding does not entail that such trials are of poor quality, it does predispose them to bias resulting from either participants' or study personnel's knowledge of intervention status (performance bias). Notwithstanding the difficulties in blinding participants and intervention providers in manual therapy trials, it is nevertheless possible to blind outcome assessors. Because lack of blinding of outcome assessors is known to result in high risk of detection bias (eg, observer bias),^{27,28} it is strongly advised that future RCTs ensure adequate assessor blinding.

Third, attrition appears to have affected both RCTs. One trial reported that 29% of controls and 14% in the active treatment group dropped out of the study.¹⁷ The second trial did not explicitly report the dropout rate of 29%, but the manuscript text suggests that the treatment outcome was recorded for only 122 participants of the 171 originally enrolled into the study.¹⁶ It is not clear, however, if these dropouts were evenly distributed between control and active intervention groups. It is well known that omitting randomized participants from the analysis in RCTs often results in biased estimates of treatment effects and that both the magnitude and direction of the resulting bias are unpredictable.²⁹ Thus, collectively, the relatively high attrition rates, uneven distribution of dropouts, and lack of intention to treat analyses suggest that the 2 trials are very prone to attrition bias.

Fourth, there are concerns about standardization of the treatment protocols. One trial simply lacked a strict treatment protocol,¹⁶ whereas in the other RCT, a relatively high proportion of the spinal manipulative interventions were directed to the upper cervical spine (28%) or other areas outside the lumbopelvic region (9%).¹⁷ Furthermore, in both trials, the diagnosis and decision regarding the active intervention (ie, spinal manipulative technique and spinal segment target) were conducted and administered by inexperienced practitioners (ie, final-year chiropractic students). It is possible that a lack of experience or technical proficiency, or both, among the treatment providers may have influenced the results in these trials.

Lastly, there are additional issues regarding lack of follow-up, adequacy of sample sizes, unexplored confounding, and possible measurement bias. In addition, the above-mentioned baseline imbalances may also have biased the statistical tests of observed differences (or lack thereof) in outcomes between active intervention and control groups.³⁰ Collectively, the issues discussed above complicate the interpretation

of the findings of the 2 RCTs and, subsequently, the conclusions that can be drawn from them.

Because the effectiveness of chiropractic care for nocturnal enuresis has not been established, it is important to acknowledge that this treatment approach can at best be described as experimental. In such circumstances, ethical practice demands that patients, or in the case of minors, their parents or legal guardians, are adequately informed before proceeding with experimental chiropractic care.

Recommendations for Future Trials

Future RCTs investigating the effectiveness of chiropractic care for nonorganic enuresis need better trial design to minimize the risk of bias. For instance, steps should be taken to ensure adequate generation and implementation of the random allocation sequence. Although it can be difficult to adequately conceal the group allocation to participants and treatment providers, outcome assessors and personnel responsible for data analysis should nevertheless be adequately blinded. Furthermore, an intention to treat analysis should be performed to ensure that the randomization is not broken and selection bias inadvertently introduced. Lastly, an *a priori* power analysis should be conducted to ensure that a trial includes the minimum sample size required to be likely to detect a clinically meaningful effect.

In addition to improved methodological quality, future trials may also consider reducing the heterogeneity of the study population and standardizing the intervention. For instance, it may be worthwhile investigating a subpopulation of patients with enuresis such as those that present with SIJ dysfunction. With such homogenous study populations, the intervention could also more easily be standardized (eg, limited to SIJ manipulation). Decisions regarding specific subpopulations and standardized interventions should be guided by clearly articulated and biologically plausible hypotheses for how the intervention might work.

Limitations

It is important to remember that causality cannot be established in case reports. That is, although chiropractic care preceded the resolution of the patient's enuresis, this does not entail that the intervention caused the resolution. Moreover, although the patient remained dry at 3 months postdischarge, it is possible that the patient relapsed later without seeking further chiropractic care. The critical review of the literature was limited by the lack of good quality clinical trials.

The findings herein should be interpreted in light of these limitations.

Conclusion

This case report described the resolution of nonorganic, primary nocturnal enuresis in an 8-year-old girl receiving side-posture HVLA manipulation of the left SIJ. A review of the current best evidence demonstrates that there is insufficient or inconclusive evidence for the effectiveness of chiropractic intervention for nocturnal enuresis.

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

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