CASE SERIES

Resolution of Chronic Nocturnal Enuresis in Children with Asperger’s Syndrome Following Subluxation Based Chiropractic Care: A Case Series

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Abstract

Objective: To describe in a case series the chiropractic care of two children with Asperger’s Syndrome suffering from long-term nocturnal enuresis.

Clinical Features: A 10-year old male, diagnosed with Asperger’s at 6 years of age, presented for care for neck pain following a fall from a trampoline. Additionally, the patient suffered from nocturnal enuresis nightly since age 2 years. Notable examination findings from static and motion palpation revealed subluxations at the C₄, T₁-T₄ vertebral body and the right sacro-iliac joint. The second patient was a 9-year-old male also diagnosed with Asperger’s Syndrome with nocturnal enuresis every night since age 5 years. Subluxations were detected at the C₅ and T₁₀ vertebral body with the sacrum restricted anterior.

Intervention and Outcome: The 10-year-old was cared for with Diversified Technique resulting in cessation of bedwetting following the first visit. The 9-year-old was cared for with Diversified Technique for 9 visits with minor improvements in bedwetting. On the 10th visit, Logan Basic was utilized resulting in cessation of bedwetting. On long-term follow-up, both patients continued to be free from nocturnal enuresis.

Conclusion: This case series provides supporting evidence on the effectiveness of chiropractic care in patients suffering from nocturnal enuresis.

Key words: Asperger’s Syndrome, nocturnal enuresis, pediatric chiropractic, vertebral subluxation, adjustment

Introduction

Nocturnal enuresis (NE) has a wide prevalence with estimates that by the age of 5 years, 15% of children will suffer from NE. The problem is twice as common in boys than girls¹³ with a self-limiting characteristic that decreases with age (for example, 15% prevalence at 5 years to 2-3% between 12-14 year olds and 1-2% incidence at ≥15-year olds).²⁴⁻⁵ NE has a spontaneous remission rate of 15% per year but as the child matures, NE persists resulting in a poorer outcome for resolution.²³,⁶ In these older children with persistent NE, 15% also suffer from encoporesis, particularly for males.²

The high prevalence of NE in the pediatric population necessitates that those involved in the care of children be cognizant of this disorder. Nocturnal enuresis is associated with a number of problems such as poor cognition, constipation, ADHD, depression and disturbed sleep. More importantly, NE places the suffering child at risk for psychological and physical abuse.⁸ In terms of conventional treatments, education and behavioral therapy, enuresis alarms and medications all have limitations.

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Case Series Narrative

Case One

A 10-year old male was presented by his parent for chiropractic consultation and possible care for neck pain following a fall from a trampoline. The patient was diagnosed with Asperger’s at 6 years of age and a history of nocturnal enuresis every night since he was 2 years old. Physical examination incorporating inspection, palpation, range of motion and neurological examination (i.e., dermatome, myotomes and deep tendon reflex testing of upper and lower extremities) was unremarkable except for the following findings consistent with vertebral subluxation. Digital motion palpation revealed the C3 vertebral body on the right side was restricted posterior, right and inferior. Digital motion palpation revealed the T1-T4 vertebral bodies were restricted posterior and the right sacro-iliac joint was restricted posterior and inferior. Based on the history and physical examination findings, radiographic examination of the cervical spine was indicated and performed (see Figures 1-3).

The radiological report interpreted a mild left inclination of the cervical spine on the A-P view with a moderate to severe anterior head carriage on the lateral view. The overall bone density appeared adequate with no evidence of fractures, dislocations or osseous pathology. A mild hypertrophy of the C7 transverse process was observed, bilaterally, that was more prominent on the left than the right.

Chiropractic adjustments were delivered using Diversified Technique\(^8\) characterized as high velocity, low amplitude thrusts. The C4 vertebral body was adjusted with the patient in the supine position. The doctor contacted the posterior aspect of the articular pillar of C4 on the right with the doctor’s right metacarpal phalangeal joint of the index finger. The T1-T4 was adjusted with the patient in the supine position. A bilateral transverse process contact was used with the doctor’s open flat hand and the supporting hand on the patients elbow. The line of drive was anterior to posterior and inferior to superior. The sacro-iliac joint was adjusted with the patient laying on their side. The doctor contacted the right sacro-iliac joint with the doctor’s left hypothenar eminence. The line of drive was posterior to anterior and inferior to superior. The patient returned the following week for a follow up visit.

According to the patient’s mother, he had completely ceased bed wetting since his last chiropractic visit. The patient continued to receive care once a week as described. During his care, the patient did not regress and continued to experience dry nights. The patient was cared for similarly with subsequent visits over a period of 10 months consisting of 18 visits. The patient was treated once a week for ten weeks, once every two weeks for four treatments and then once a month for four months. Follow-up over two years later revealed the patient as free of bed-wetting. The patient’s mother indicated to the attending chiropractor that she could not express fully how this has made such a difference in her son’s self-esteem and life. According to the patient’s mother, her son “has been lifted to new heights and is now looking forward to sleepovers without fear of embarrassment.”

Case Two

Given the results of care provided in the patient described above, the patient’s mother referred another patient to the attending chiropractor with a similar problem of nocturnal enuresis. The second patient was a 9-year-old male medically diagnosed with Asperger’s Syndrome. This 9-year-old male suffered from nocturnal enuresis every night since he was 5 years old.

Physical examination (i.e., inspection, palpation, range of motion and neurological examination) was unremarkable except for the following findings consistent with vertebral subluxation. On digital palpation incorporating motion palpation, the C5 vertebral body was restricted posterior, right and inferior. The T4 vertebral body was restricted posterior and the sacrum was restricted anterior. The patient’s mother consented to a trial of care of once per week with the child receiving chiropractic care characterized as high velocity, low amplitude thrust type adjustments (i.e., Diversified Technique\(^8\)).

The patient’s response to care was experiencing an occasional dry night throughout the week, but the bedwetting soon thereafter resumed at a frequency of 7 nights per week. The patient was adjusted once a week for 9 weeks with intermittent results of dry nights. On the 10th visit, the attending chiropractor decided to change her adjusting technique to Logan Basic.\(^9\) The patient was observed to be hyperactive and the chiropractor wanted to focus on his parasympathetic nervous system.

While a thumb contact on the left sacro-tuberos ligament and a left thumb contact on his left sub-occipital area was applied, the patient was observed to have a noticeable slowing down of his breathing. The contact was held for 1-2 minutes. No other segments were adjusted thereafter at this visit. The patient seemed very calm and relaxed following this visit. With Logan Basic as the mode of chiropractic care, the patient stopped wetting the bed thereafter. The patient was cared for as described above with subsequent visits over a period of 22 weeks consisting of 22 visits. These visits were qualified in part, to other benefits the patient was receiving such as improved demeanor and more calm. Two years follow up revealed that with the change in chiropractic technique, the patient continued to experience dry nights thereafter.
The estimated prevalence of NE is highly variable due to great heterogeneity in the diagnostic criteria of the disorder. The different types of NE\textsuperscript{20} are summarized in Figure 4. The variants of NE can be gleaned from the history examination and should focus on the timing, frequency, volumes and duration of symptoms to arrive at a diagnosis of NE or its subtypes.\textsuperscript{21} For example, if there is a history of daytime voiding, this may be due to dysfunctional voiding (i.e., LUTS) or urinary incontinence. If the child has a history of dry nights >6 months, the NE (i.e., secondary MNE) may be due to a psychological problem or stress such as bullying or change in living conditions/geography, divorce, etc. A query on the volume of fluid intake prior to going to bed may suggest nocturnal polyuria, diabetes mellitus or insipidus. Weight loss or fatigue may direct to diabetes or kidney disease or something as a family history of NE may suggest a genetic component. Note that there is an association of constipation with NE.

In terms of the physical examination, the attending chiropractor should be cognizant of the differentiating factors for functional versus anatomical causes of NE.\textsuperscript{21} For example, note the child’s growth pattern as poor growth may indicate renal disease. Abnormalities of the lumbosacral spine are associated with spinal dysraphism while abnormal neurological findings with the perineum and the lower extremities is indicative of spinal cord abnormality. All these may manifest as NE. Slow urinary stream or dribbling may indicate urethral abnormality.

The pathogenesis of NE is summarized in Table 1.\textsuperscript{21} Of interest in our examination of the literature is the recent support for orthodontists to optimize treatment for NE. Juszczak\textsuperscript{22} argued that dental maxillary expansion improves breathing and oxygenation due to the increased nasal and nasopharyngeal airway dimensions. Second, improved nasal breathing results in reduced apnoic episodes leading to better lymphatic circulation and increases the antidiuretic function of the pituitary gland.

Third, there is improved breathing capacity and facilitates improved oxygen saturation, which may be beneficial for sleeping and facilitate children to wake up more easily and detect bladder fullness. Fourth, sufficient blood oxygenation may result in improved neuromuscular coordination and control of bladder sphincter during sleep. These arguments are reminiscent of Goodheart’s advice to practitioners to look not at addressing the lumbar spine but to the C\textsubscript{3} vertebral level.\textsuperscript{23}

According to Goodheart, this is a physical, functional, structural problem associated with disturbances of the segments, not at the kidney and bladder areas of the spine, but at C\textsubscript{3}, which is associated with the innervations of the phrenic and intercostals nerves. The phrenic nerves supply motor fibers to the diaphragm (the muscle for respiration) and sensory fibers to the fibrous pericardium, mediastinal pleura, and diaphragmatic peritoneum. The intercostal nerves supply motor fibers to the thorax and abdomen for contraction of muscles as well specific sensory fibers from the skin and parietal pleura. If the process of breathing is affected, high concentrations of CO\textsubscript{2} inhibits smooth muscle contraction.\textsuperscript{24} The detrusor muscles are smooth muscle layered within the bladder and arranged in spiral, longitudinal, and circular bundles. When the bladder is stretched, signals from the parasympathetic nervous system are sent to contract the detrusor muscles. This encourages the bladder to expel urine through the urethra. With high CO\textsubscript{2} concentrations, smooth muscles such as the detrusor muscles may not be functioning. In addition to the above advice, Goodheart commented that fluid restriction and interruption of the child’s sleep by the parent to allow the child to void any accumulation of fluids is good practice.

**Chiropractic Care**

Review articles have examined chiropractic care for a number of pediatric conditions.\textsuperscript{25-26} Specific to nocturnal enuresis, Bronfort et al.\textsuperscript{25} concluded that there is inconclusive evidence in a favorable direction regarding the effectiveness of chiropractic care for the treatment of nocturnal enuresis.\textsuperscript{27-28} Clar et al.\textsuperscript{26} essentially arrived at the same conclusion. When one examines the studies thus far published beyond case reports on the care of patients with NE, the literature is disappointing.

LeBoeuf et al.\textsuperscript{29} examined a cohort of 171 children with NE. For a period of two weeks, their rates of nocturnal enuresis were measured as initial outcome measures. Following the 2-week period, the children were then placed into two groups: Group A (N=71) receiving no care for a period of two weeks and Group B (N=100) where the children received chiropractic care. The chiropractic technique utilized was characterized as Diversified Technique involving high-velocity, low amplitude type thrusts. Care was halted when the children attained 2 wet nights in a two-week period. The median number of wet nights per week was 7.0 at the onset of the study. After 2 weeks without any therapy, the number of wet nights had decreased to 5.6 and by the end of the treatment this number was 4.0.

Following the course of treatment, 15.5% of subjects wet a maximum of 2 nights per fortnight, or, where data for the last 2 weeks of therapy were unavailable, a maximum of 1 night/week. According to the investigators, their results were less favorable than the therapeutic success of other common types of therapy. The only variable, which predicted treatment outcome, was the initial estimate of bed-wetting; the more severe the condition at the onset, the less likely was the child to improve by the end of the study.

A major criticism of the Leboeuf et al. study includes the use of chiropractic interns as opposed to experienced chiropractors in the care of the children. In addition, some question the appropriateness (i.e., too short a period of time) of the period of time used to measure the baseline wetting frequency as well as the post-treatment period. Furthermore, the description of the examination procedures employed and the type of care applied are not well described. Reed et al.\textsuperscript{30}, examined 57 children randomized into two groups: an SMT group receiving the Palmer Package (i.e., a combination of techniques) and Sham group (i.e., Activator setting at “0”).

Prior to providing care, a baseline measure was performed for two weeks. The children were then cared for in their
respective groups for a period of 10 weeks. Outcome measures were then performed for another 2 weeks followed by exit interviews of all patients. The number of wet nights for the control group was essentially unchanged. The treatment group had an approximate 18% decrease in the number of wet nights per 2 weeks. According to the investigators, the mean pre-treatment and comparative change in the wet night frequency for the treatment group compared with the control group did not reach statistical significance. However, 25% of the treatment-group children had a 50% or more reduction in the wet night frequency from baseline to post-treatment while none among the control group had such reduction. Van Poecke et al. described 33 consecutive patients attending care at one chiropractic clinic over a 3-year period, for PNE. The care provided followed the NeuroImpulse Protocol.

All patient records were analyzed for the number of wet night frequency at baseline and at 3, 6, 9, and 12 months comparative. In addition, the number of treatment visits over a 12-month period was noted and the presence of constipation and/or positive family history at presentation. Data were analyzed using descriptive statistics, Friedman’s test, and Dunn’s Multiple Comparison test. Of the 33 patient records analyzed, 22 showed resolution of PNE during the 12 months following initiation of chiropractic care. The mean number of visits in the responders group was 2.05 +/- 1.33. Ten responders presented with constipation and a further 8 with a positive family history of PNE. Resolution of constipation was noted to be essential to the successful response to treatment as this condition and positive family history of NE were poor prognostic factors.

Given these findings, we encourage continued reporting on the success and failures of chiropractic care in the care of children with nocturnal enuresis. An examination of previous case reports published on the success of chiropractic care in children enuresis reveals that to date, most of the studies performed on the pediatric population involved the use of the HVLA type thrust spinal adjustments. Alcantara et al. lamented the limitations on the use of this type of spinal adjustment in the asthma clinical trials. As we showcased in this case series, we opened for the possibility that not all children may respond to HVLA type care and that other types of chiropractic maneuvers/techniques may be effective and must be investigated. As such we must examine the aforementioned systematic reviews thus far with caution and to use an idiomatic expression, “not throw out the baby with the bath water.”

Electroencephalography (EEG) studies in individuals with NE have reported cortical dysmaturity. Khedr et al. examined 41 patients with NE using using transcranial magnetic stimulation (TMS). Each subject was assessed clinically regarding frequency, duration of their enuresis along with a Health Survey Measurement. Neurophysiological measures included resting and active motor thresholds (RMT, AMT), motor evoked potentials (MEP) of the upper and lower limbs, cortical silent period duration (CSP) and transcallosal inhibition (TCI), in the upper limbs. The investigators found that NE patients had a significantly lower Health Survey Measurement score for both physical and mental health components compared to the control group. RMT and AMT of both upper and lower limbs as well as the duration of the CSP and TCI were significantly reduced compared with the control group. There was significant positive correlation between RMT, AMT and Health Survey Measurement scores, especially Social Functioning. Based on these findings, the authors concluded that patients with nocturnal enuresis are characterized by pathologically increased excitability and reduced inhibitory processing in the motor cortex, which could contribute to the pathogenesis of nocturnal enuresis. In light of these findings, we are reminded of the studies by Taylor and Murphy demonstrating altered specific central corticomotor facilitatory and inhibitory neural processing and cortical motor control or altered cortical integration of dual somatosensory input following adjustments to the cervical spine. Dishman et al. demonstrated that amplitudes of the motor-evoked potentials were significantly facilitated from 20 to 60 seconds relative to the pre-baseline value following L5-S1 spinal manipulation. Further research is required to elucidate the true nature of the chiropractic adjustment resulting in neurological changes to benefit patients suffering from NE.

Von Gantard commented that childhood conditions such as nocturnal enuresis, daytime urinary incontinence, lower urinary tract symptoms and fecal incontinence are more common in children with special needs than in typically developing children. Our study thus far is the second reporting in the scientific literature describing the successful care of a special needs child and the first in children with Asperger’s Syndrome suffering from NE. Noriega described a 6-year-old boy with Autism and cared for with upper cervical technique (i.e., NUCCA). We encourage more reporting on the benefits of chiropractic care for this pediatric population.

A post-positivist perspective is based on the ontology of materialism and objectivity is the key to understanding the world around us. Based on this perspective, we caution the reader on the generalization of the case reported due to the presence of bias. These biases include the lack of a control group, spontaneous remission, self-limiting course and natural history of the disorder, subjective validation, and expectations for clinical resolution on the part of the patient. However, from a constructivist perspective where perception creates reality and honors the individual experience, we would argue that the success of this case report forms the basis for our generalization in caring for similar patients.

Conclusion

This case report highlights that patients suffering from nocturnal enuresis may benefit from chiropractic care and highlights the need to explore multiple chiropractic techniques and their effectiveness in similar patients. We support continued research in this area of chiropractic care.

References


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<thead>
<tr>
<th>Nocturnal polyuria</th>
<th>Nocturnal polyuria refers to the increased production of urine during asleep. This may be as the result of increased fluid intake prior to sleep and/or the effects of antidiuretic hormone (ADH) due to decreased secretion or resistance to the hormone. In addition to decreasing urine production, ADH has also been shown to aid in increased bladder distention.</th>
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<td>Decreased bladder capacity</td>
<td>Despite findings that no anatomical differences in bladder size have been observed in children with or without NE, some studies have found a reduction in nocturnal bladder function, as opposed to daytime function. Additionally, its been found that in children with NE, their nighttime voided volumes are significantly decreased compared to daytime voided volumes.</td>
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<td>Detrusor over-activity</td>
<td>This is based on the theory that children with NE have a lower pressure threshold for detrusor activity due to the finding that children with NE have significantly decreased functional bladder capacities compared to healthy age-matched children.</td>
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<td>Associated sleep arousal disturbances</td>
<td>This is based on the theory that children with NE have basic discordance in the ability to arouse from sleep from voiding urge sensation. Whether this is caused by sleep disturbances or problems with the bladder-brain communication remains unknown. Studies have demonstrated that children with NE have detrusor instability while asleep, but not while awake and children with NE tend to be “deeper sleepers” than other children and children with NE to be “light sleepers” or no different than non-NE children.</td>
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<tr>
<td>Bladder brain connection dysfunction</td>
<td>This is based on the theory that a reduced ability to inhibit micturition during sleep may originate from a dysfunctional pontine tegmentum. Children with NE demonstrated less inhibition of their blink response as controlled by the pedunculopontine tegmental nucleus, which is located near the pontine micturition center in the brainstem. When treated, sleep arousals and inhibitory responses improved in children with NE.</td>
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<td>Global maturation delay</td>
<td>It’s been theorized that that global CNS maturation delay may be a contributor to NE. In longitudinal studies, children with NE have been found to have progressive maturation of bladder stability in conjunction with EEG findings that suggest increased CNS recognition of bladder fullness and the ability to suppress the onset of bladder contraction.</td>
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Table 1 The pathogenesis of nocturnal enuresis.²¹
Figure 1. Anteroposterior view of cervical spine

Figure 2. Anteroposterior open mouth (APOM) view of the cervical spine
Figure 3. Lateral view of the cervical spine

Figure 4. The different types of nocturnal enuresis

Nocturnal Enuresis (NE):
Intermittent incontinence during sleep in children >5 years of age despite acquired sphincter control.

Monosymptomatic NE (MNE):
Nighttime bedwetting in the absence of any other lower urinary tract symptom (LUTS), and absent history of bladder dysfunction.

Non-monosymptomatic NE:
NE in children with other LUTS and has a positive history of bladder dysfunction.

Primary MNE:
A child with NE without a history of nocturnal urinary incontinence (UC) and has never experienced dry nights.

Secondary MNE:
Children with nocturnal UC for at least 6 months and experience NE secondary to psychological stress or an organic cause.