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JCCP JOURNAL OF CLINICAL CHIROPRACTIC PEDIATRICS

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GUIDELINES FOR AUTHORS

JCCP JOURNAL OF CLINICAL CHIROPRACTIC PEDIATRICS

The *Journal of Clinical Chiropractic Pediatrics* welcomes original and scholarly manuscripts for peer-review and consideration for publication. Topics must pertain to the field of pediatrics which includes pregnancy and adolescence. Manuscripts should not have been published before or submitted to another publication.

The following will be considered:

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Pilot Studies or Hypothesis – papers which, while very broad, present with a clear hypotheses and suggest a foundation for future, in-depth studies.

Literature Reviews – studies of existing papers and books presented with the intention of supporting and encouraging new and continuing study.

Technical Descriptions – reports of new analytical/diagnostic tools for assessment and delivery of care. Controlled, Large Scale Studies – usually, but not necessarily, performed at a college or research facility. May be double-blinded.

Commentaries – presentations of opinion on trends within the profession or current events, pertaining to pediatric and adolescent chiropractic care.

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5. Consent form for photographs (if applicable)
6. Letters of permission to use previously published materials (if applicable).
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A Short History of Chiropractic Pediatric Education

In 1991, a movement was started to provide education in quality chiropractic pediatric and maternal health care. That was the year that the International Chiropractors Association (ICA) organized and held its first pediatric conference in San Diego, California. With more than 500 doctors in attendance, enthusiasm soon developed for the formation of a more formal pediatric post-graduate education program.

In 1992, the Council on Chiropractic Pediatrics was established under the auspices of the International Chiropractors Association. The Council on Chiropractic Pediatrics' three-year post-graduate education program was developed by a small group of practitioners with academic and teaching experience that included Maxine McMullen, Joan Fallon and myself.

The program was first presented in Chicago in 1993 with the sponsorship of Palmer College of Chiropractic and the first class graduated in Chicago in May 1996 with a group of 27 doctors earning board certification and the DICCP credential (Diplomate in Clinical Chiropractic Pediatrics) from the International Chiropractors Association Council on Chiropractic Pediatrics.

The DICCP board certification program has been presented in United States, Canada, Australia and New Zealand with support and assistance from chiropractic colleges in those countries. At the time of writing this editorial, 240 chiropractors have been board certified at the end of their 3 years of post-graduate study.

The Journal of Clinical Chiropractic Pediatrics (JCCP)

In 1995 the need was recognized for a peer-reviewed journal to showcase the work of the many chiropractic clinicians, educators and researchers dedicated to the advancement of quality pediatric and maternal care. An editorial board was established and the first issue of the *Journal of Clinical Chiropractic Pediatrics* (JCCP) was published in January 1996. Publication has continued since that time.

JCCP continues to provide quality articles under the editorial guidance of Drs. Sharon Vallone and Cheryl Hawk. Since 2014, JCCP has been published as an open-access online journal available, free of charge, to a wide audience of interested readers. JCCP is committed to publishing research, scientific and professional papers, literature

reviews, case reports and clinical commentaries relevant to the health and treatment of the pregnant, postpartum and pediatric patient. Articles are received for publication from around the world, making this a truly international review of the work being carried out in many countries.

The year 2016, a momentous year for Chiropractic Pediatrics

In December 2016 at a Conference in Hawaii, the ICA Council on Chiropractic Pediatrics celebrated 25 years of pediatric education it has organized and presented in countries around the world. Special thanks to Molly Rangnath for having expertly guided all facets of this program since its inception.

Chiropractic care for children is increasingly becoming a vital part of a family's health care program. The educational opportunities and access to pediatric research provided by the ICA Council on Chiropractic Pediatrics and the *Journal of Clinical Chiropractic Pediatrics* offer doctors of chiropractic the knowledge and tools they need to meet the challenges of family practice with a greater level of skill and confidence.

I believe it is both a privilege and duty of our profession to care for the health of our children and to continue to advance knowledge and research in the field of chiropractic pediatrics.

To the doctors who have dedicated their professional careers to the advancement of chiropractic care for the pediatric patient, we applaud and thank you.

Some time ago, someone gave me an inspirational plaque that sits on my desk to this day. It reads:

"A hundred years from now it will not matter what your bank account was, the sort of house you lived in, or the kind of car you drove... But the world may be different because you were important in the life of a child."



Peter Fysh, DC, FICCP
Professor Emeritus, Palmer College of Chiropractic West

Chiropractic care for the cervical spine as a treatment for plagiocephaly: a prospective cohort study

By Nicola Ann Douglas, MChiro, MSc¹, Maria Browning, BSc, DC, MSc (Chiro Paeds), Cert Med¹, Joyce Miller, BS, DC, PhD²

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ABSTRACT

Background: Plagiocephaly is a condition that affects the shape of the skull in infants. The research has suggested that there is a growing association between plagiocephaly and developmental delay later in infancy extending into childhood. Plagiocephaly is an increasingly common condition in society which often presents to a chiropractic practice. There have been no studies prospectively investigating the outcome of chiropractic care on a group of infants with plagiocephaly. **Objectives:** To observe any change in head deformation measurements in a single cohort group of infants aged 0-12 months old presenting to a chiropractic clinic and receiving chiropractic care over a course of six weeks. **Setting:** This single cohort observational study took place at a chiropractic teaching clinic between February and July 2015, on the south coast of England. **Methods:** Infants presenting to the chiropractic clinic with the complaint of plagiocephaly were measured from the external occipital protuberance (EOP) to the anterior ear both right and left sides during their routine course of treatment. These measurements were re-examined at the end of their course of care or at six weeks after presentation, whichever ever occurred first. **Results:** A total of 64 infants were included. The mean change in the plagiocephaly measurement was a reduction of $1.13\text{cm} \pm 0.89\text{cm}$, $p = 0.00$, 95%CI (-1.36 to -0.92cm). Overall, 20 out of the 64 participants showed a full resolution of plagiocephaly, with a final measurement below 0.4cm difference in occipital measurement side to side, which is considered normal. No adverse events were reported for any of the infants. **Conclusions:** Overall, there was both a statistically and clinically significant reduction in plagiocephaly measurement for this cohort of infants after a course of chiropractic care. As this was an observational study, this cannot be interpreted as cause and effect. However, these results encourage further research, particularly a RCT to investigate the effect of chiropractic care on plagiocephaly in infants.

Introduction

Plagiocephaly is a term used to describe an asymmetry in the shape of the skull. Non-synostotic plagiocephaly describes a flattening of the skull that evolves into two types of shape deformities (Figure 1). Firstly, the posterior-lateral

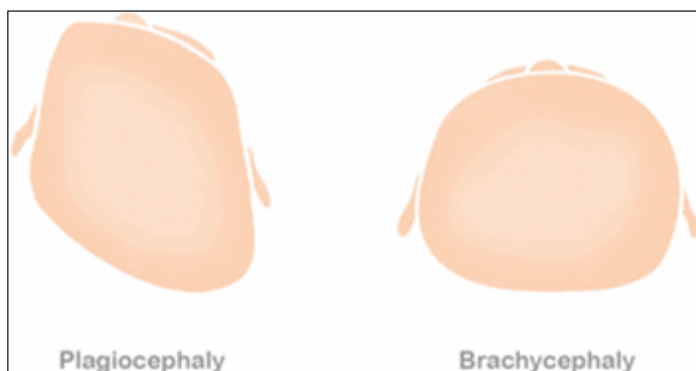


Figure 1: A comparison of the two different head asymmetries in positional plagiocephaly, amended with permission from Saich and Laker 2014.³⁷

occiput can appear flattened, termed plagiocephaly and secondly a uniform flattening across the back of the occiput is termed brachycephaly.¹

The cause of plagiocephaly is exposure of the cranium to external compressive forces while it is still developing. The flatness is usually first recognised by parents when the infant is 2-3 months old.² Since the 'Back to Sleep Program' in 1992 there has been a significant rise in the prevalence of plagiocephaly. However the actual prevalence of plagiocephaly is uncertain due to the range in estimates. Estimates vary from 16-21% at 6-7 weeks old and 20% at 4 months old³ to 47% of infants aged 7-12 weeks old out of cohorts of healthy full term infants.⁴ The real issue is not only the rise in incidence but also long-term risk factors. The UK National Health Service (NHS) states plagiocephaly is "cosmetic only."⁵ This statement contradicts the research suggesting that plagiocephaly is associated with learning disabilities, dysfunctional auditory processing and developmental delay in the gross motor and cognitive domains.^{6,7,8,9,10}

There is further conflict of opinion as to whether plagiocephaly naturally resolves as the head develops or whether interventions may be helpful to change the long-term course of the condition.^{10,11} There have been several studies comparing the effects of cranial orthosis against physical therapy or active counter positioning therapy on plagiocephaly in infants. However, there is no evidence of benefit for cranial orthoses or helmets in the treatment of plagiocephaly.¹

The research suggests that physical therapy or active counter positioning may be effective when the infants are young with mild to moderate plagiocephaly.¹² One randomized controlled trial (RCT) found active counter positioning advice to parents/guardians had a positive and significant effect on plagiocephaly in infants compared to no instruction.¹³ It is inconclusive whether physical or manual therapy may be more important than what is currently understood for the appropriate management of plagiocephaly which is no clinical management. The purpose of this investigation was to assess whether there is a role for chiropractic manual therapy in the routine treatment of plagiocephaly in a clinical population.

Background

There is little research investigating the effect of chiropractic care, a form of manual therapy, on plagiocephaly. There has been one retrospective study¹⁴ and several case studies.^{15,16,17,18,19} Also there has been one pilot single cohort study investigating the effect of osteopathy, another form of manual therapy on plagiocephaly in infants.²⁰ All of these studies showed improvement in plagiocephaly in infants.

Chiropractors commonly manage infants with plagiocephaly and treat the cervical spine to achieve full range and freedom of motion so that the infant is not “stuck” in one position. If the head can be freely moved, then the pressure on the cranium will be equally distributed and the skull will, ideally, become symmetrical.

This is an appropriate method for resolution as research has shown there is a relationship between cervical range of motion and plagiocephaly.^{7,21,22} Decreased cervical motion was found to be associated with a larger cephalic index (calculated by cranial width divided by cranial length, multiplied by 100) and brachycephaly.²¹ It seems intuitive, but research has also found that infants with head positional preference are four times more likely to have plagiocephaly than infants without positional preference.²² Their study found that the condition is at increased risk when infants do not vary their head position and leads to one question: is this because they are unable to vary their head position? This tendency has also led to referring to plagiocephaly as posi-

tional head deformation (PHD). We found this easier to say and understand by the parents as well as more descriptive of the condition and we use that term clinically.

The link of head deformation to suboptimal cervical spine rotation has been corroborated with research that has shown that physical therapy improved cervical range of motion in infants with plagiocephaly and this significantly improved cranial symmetry compared to a control group receiving counter positioning advice only.²³ Based on those findings it was recommended to general practitioners to consider manual therapy for the early management of infants with plagiocephaly.²⁴ Despite the fact that chiropractors are known for manual therapy and are the most common choice for families choosing CAM care for their infant,²⁵ there is still a great need for further research investigating the outcomes of chiropractic care with positional head deformation prospectively in a clinical setting.

Method

This study was a prospective cohort study conducted at the Anglo-European College of Chiropractic (AECC) clinic, Bournemouth, UK. The AECC ethics subcommittee granted ethical approval in January 2015.

A literature search was conducted using the online databases of Cochrane Library, Index to Chiropractic Literature, Pubmed and Science Direct. Key words were ‘deformational plagiocephaly,’ ‘positional plagiocephaly’ and combined individually and together with ‘chiropractic,’ ‘infant’ and ‘manual therapy.’ Publications of RCTs, systematic reviews, case studies, cohort retrospective and prospective studies were included. Publications involving craniosynostosis or surgical cases were excluded as well as publications before 2001 to allow for the most current literature. Thirty-one articles were found and obtained through the online and library resources at Bournemouth University and were reviewed. A cross-reference search was also conducted for any relevant but unfound studies at the end of each paper. Several studies were relevant but were all included in the databases search. A hand search for relevant but unpublished studies and this was also conducted at Bournemouth University; one thesis was found.

Population

The sample group of participants were infants aged between 0-12 months old, diagnosed with plagiocephaly whose parents presented their child to the AECC clinic for care and gave their consent for the infant to receive chiropractic treatment. Participant data was excluded from this observational study if the infant was diagnosed with craniosynostosis, or undergoing any other form of medical treatment or therapy.

Procedure

The routine chiropractic examination at the AECC clinic involves a musculoskeletal assessment recording head shape as well as other key measurements such as vital signs documented in each infant’s file. Positional Head Deformation (PHD) was determined by two cranial measurements taken from the external occipital protuberance (EOP) to the anterior ear for both left and right side (Image 1). A difference of more than 0.4cm between the two sides was recorded as plagiocephaly or PHD present.²¹ These measurements were taken using the prototype occipital measurement device (Image 2) that has shown to have a high inter-examiner reliability.²⁶ Following the standard AECC clinic’s protocol for treatment of infants and children, these measurements were taken at the 1st, 4th and 7th treatment visit, and/or at the date the child was released from care or monitored over a maximum time frame of six weeks. Six weeks was chosen because reduction in that time frame would be considered well ahead of the natural history of the disorder.



Image 1: A photograph of measuring the infant’s EOP to anterior ear using the prototype occipital measuring device.



Image 2: The prototype occipital measuring device.

Following a routine course of chiropractic care for infants with plagiocephaly, the infants’ data were gathered from each of their files. Confidentiality for each participant remained intact throughout the study which was purely observational as the condition is known to have long-term negative sequelae. There was no control or comparison group that did not receive care, as this could be considered unethical in the light of the negative long-term prospects associated with the condition.^{6,7,8,9,10}

Results

A total of 64 participants met the inclusion criteria. The clinical characteristics of the participants are shown in Table 1. There were more males than females, 62.5% (n=40) and 37.5% (n=24) respectively. The age range of infants presenting to the clinic varied from 2 to 44 weeks old, average age was 11.5 ±6.76 weeks old, 95%CI (9.84 to 13.2weeks). 63 of the 64 participants were aged under 6 months with the exception of one participant aged 44 weeks (11 months) old.

Table 1. The Clinical Characteristics of the Infant Participants in Plagiocephaly Study

		n	Percentage
Gender	Male	40	62.5
	Female	24	37.5
Age presenting to clinic	0-7 weeks	19	30
	8-15 weeks	32	50
	16-23 weeks	10	16
	24-52 weeks	3	4
Feeding	Breast fed	23	36
	Bottle fed	28	44
	Both	13	20
Gestation Length	Full term	23	76
	Pre-term (≥37weeks)	7	24
Type of birth	Natural	13	44
	Caesarean Section	10	33
	Assisted	7	23
Child Number	Firstborn	22	73
	Sibling	8	27
Multiple births	Twins	7	23
	Single	23	77

This study found that there were differences in the record keeping so there were different numbers who answered specific questions. The number (n) is shown in the tables, along with percentage. On average, most infants were full-term with 56% assisted births and 44% natural vaginal deliveries. All of the infants had a limitation in cervical spine range of motion, with 46% having preference to rotate to the right, 40% to the left and 4% where parents did not know the direction of their child’s preference.

The average difference in side-to-side EOP to anterior ear

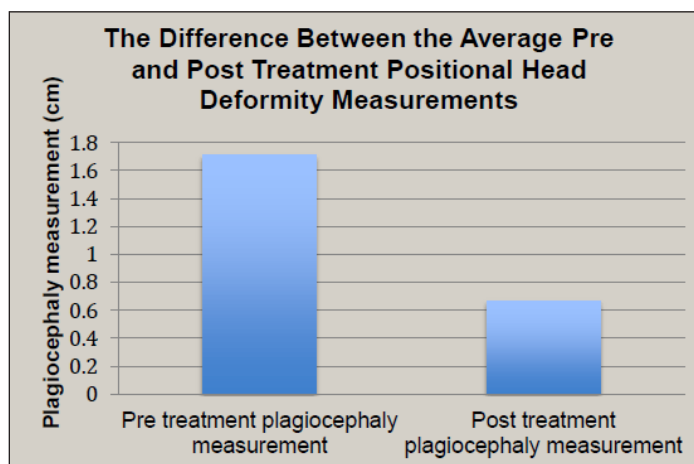


Figure 2: Plagiocephaly measurements before and after chiropractic treatment.

measurement for this group of infants before chiropractic treatment was 1.71 ± 0.84 cm, 95%CI (1.50 to 1.91cm). At the end of the course of chiropractic care these measurements reduced to 0.67 ± 0.65 cm, 95%CI (0.50 to 0.83cm), (Figure 2). The mean change in the occipital measurement was a reduction of 1.13 cm \pm 0.89 cm, $p=0.000$, 95%CI (-1.36 to -0.92cm), on average a 33% decrease in difference side to side. Overall, 20 out of the 64 participants showed a full resolution of head deformation with a measurement difference below 0.4cm.

All participants ($n=64$) had full range of cervical motion restored following the course of chiropractic care. On average, the course of chiropractic care consisted of five treatments over a period of six weeks. The infant's head circumferences changed throughout the course of chiropractic care as one would expect with growth. The average head circumference for 21 participants in this study's sample of infants at the start of the study was 40.6 cm \pm 2.10 , 95%CI (39.7 to 41.5cm) that increased to 42.2 cm \pm 2.56 , 95%CI (41.1 to 43.3cm) at the end of the study. Overall there was a mean increase of 1.61 cm \pm 1.68 , 95%CI (0.89 to 2.33cm).

Discussion

This study was designed to observe infants presented for care for positional head deformation by their parents over a course of chiropractic treatment lasting up to six weeks.

This study's population was representative of infants with PHD. For example there were more boys than girls. Plagiocephaly is more prevalent in males.² However, another recent study found plagiocephaly was present in an equal number of males and females in a healthy cohort of neonates²¹ and this is likely due to the general increased prevalence of plagiocephaly.⁴ It must be noted that the Aarivala et.al. 2014 study included only neonates and perhaps it is that both genders have equal amounts of head deformation as newborns but that it is more likely to persist in males. This concept has never been studied.

Also, the majority of the participants were under six months (98% $n=63$) with half between 2-4 months old; other studies report that plagiocephaly is most prevalent between 2-4 months old.^{1,27,28} One explanation is due to the acceleration in the infant's head development at this age. Any excessive compression to one aspect of the skull will alter the shape and appear as flattened in comparison to the rest of the skull's normal development.²¹

Another example that this study's population was representative of the general PHD population, was a quarter of the subjects had been born prematurely, which is higher than the average incidence in developed countries of 5-9%.²⁹ Other research suggests that prematurity is over-represent-

ed in children with misshapen heads.^{28,30}

The same is true for over-representation of assisted births which accounted for over half of the births in this cohort. The average in the UK population is 39%.³¹ Plagiocephaly is associated with assisted births.²¹

Almost $\frac{3}{4}$ of this sample were first borns and $\frac{1}{4}$ were twins, far outnumbering averages for routine births in the UK population (equal ratios between first born, second born and third or more born in the UK³² and 1.52% of all births in the UK were multiple births of twins in 2012).³³

Cervical mobility restrictions were reported in all of the infants in this sample. Research has suggested when the infant's neck has limited movement, the infant will have a preference for a particular side to turn as they are unable to turn equally to both right and left.^{7,21} These findings also support research that cervical restriction and plagiocephaly are linked with neurological disruption and developmental delay due to factors associated with inactivity and variable tone.^{7,9,34} It is unlikely that the association with developmental delay is due to the head deformation, but more likely to be related to the poor movement patterns associated with it. This highlights the need for manual therapy to restore normal range of motion and beneficial movement.

The infants' plagiocephaly measurement significantly reduced over the course of chiropractic care. Out of the sample of 64 participants, 20 showed a complete resolution of head deformation, a value of 0.4cm or less difference between left and right side which is widely recorded as a normal, almost imperceptible difference.² The remaining infants showed a reduction in the plagiocephaly measurement with a mean reduction of 1.13 cm \pm 0.89 cm, $p=0.000$, 95%CI (-1.36 to -0.92cm). That improvement was both statistically and clinically significant and shows a percentage decrease of one third and the improvement would be expected to continue with head growth, with the impediment to cervical range of motion removed.

Chiropractic treatment involved pediatric manipulative therapy to the spine and extremities through the form of press and hold methods aimed to mobilize any joints involved, a low force treatment of 2-8 newtons depending on the age and condition of the child. The chiropractor also gave advice to parents on supervised prone playing (tummy time) for their infant, as well as home measures for active counter positioning to encourage the child to look to all directions. Although there are no clear updated guidelines on prone play (tummy time) for infants, the AAP (1998)³⁵ did suggest that it was important to start the first week with awake-time repositioning as the supine sleep program had resulted in an epidemic of head shape problems. There

were no negative side effects or adverse events reported during the course of chiropractic care.

There are limitations to this type of study. Not only is there not a control group for comparison but this is a time period when infant's heads undergo accelerated growth. In fact, this factor most likely contributes to the rapid improvement in head shape. The average head circumference is 35 cm in full term neonates increasing to 45 cm by 1 year old and 55cm in a grown adult.³⁶ Therefore, there is rapid head growth within the first year of the infant's life. Head circumference increases, on average, by 2cm per month for the first 3 months of life, 1 cm per month for the second 3 months of life and 0.5cm per month from 6-12 months.³⁶ However, there is little evidence that head shapes change along with growth unless the infant is able to perform full cervical range of motion and compress both sides of the occiput evenly. It does seem as though a key to improvement is to facilitate full cervical range of motion, which is what the chiropractic care was designed to do.

That said, the plagiocephaly measurement could have reduced naturally given significantly more time. It has been discussed in previous studies that infant's head shapes return to their normal shape by 3-5 years of age.^{1,11,13} Other research has found contrary findings and suggested plagiocephaly remains further into childhood.¹⁰ Therefore the reduction in plagiocephaly seen amongst this sample of infants could have been down to their natural head development. However the research has suggested this process occurs later in infancy and early childhood instead of during early infancy. It can be said that the changes in this study have occurred ahead of the natural history of the disorder.

Further, all infants received the full spectrum of chiropractic care which involved manual therapy, along with advice for counter positioning and tummy time. Therefore, the positive effects found after chiropractic care could have been the effect of counter positioning and tummy time measures given by the parents instead of the application of chiropractic manual therapy. Counter positioning measures have been found to have positive outcomes of a reduction in plagiocephaly measurements in infants when compared to cranial orthosis¹² and as a measure on its own.¹³ However, research has also shown that compared to physical therapy, greater effects were seen in the physical therapy group²³ in two groups of infants with plagiocephaly. There have been no studies prospectively investigating the effect of chiropractic care on plagiocephaly in infants compared to parent measures. In future studies it would be interesting to compare the two therapeutic measures and have two groups of infants with plagiocephaly, one group receiving chiropractic care and one counter positioning techniques. However, ethical consideration is required with this type

of study and since there is some evidence that both work, it may not be ethical to withhold any type of effective treatment, considering the long term negative associations with unresolved plagiocephaly.

Another limitation to this study was the prototype measurement device used to determine the plagiocephaly value. This has been shown to have high inter examiner reliability²⁶ but it has not been validated. There is also the limitation of human error and the clinicians using this device may have not correctly read the infants' EOP to anterior ear measurement. This prototype measurement device was used as it was easy and inexpensive to implement in a routine clinical setting compared to other anthropometric or digital measuring devices. However, by choosing this device it does have its' limitations such as the measurement from EOP to anterior ear difference between left and right side would only show one form of plagiocephaly, positional plagiocephaly instead of brachycephaly as well. Also the prototype measurement device is not a standardized device for measuring plagiocephaly, therefore this study is more difficult to compare to other studies using different devices. However, parental perception of improvement is at this time the gold standard for outcomes, and this device compared favourably to simple observation by parents and clinicians as well as supplying some level of objectivity.

Conclusion

This study showed a significant reduction in head deformity in infants during a course of six weeks of chiropractic care. Although this cannot be interpreted as cause and effect because the research was observational in a routine clinic setting rather than a randomized controlled trial, the improvements in head shape occurred earlier than the natural course of the condition. Further research is strongly encouraged to investigate the effects of chiropractic management for this condition.

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Clinical effects of spinal manipulation in the management of children and young adults diagnosed with autism spectrum disorder — a systematic review of the literature

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ABSTRACT

Background: Autism spectrum disorders (ASD) are classified as pervasive developmental disorders that permanently affect essential mental functions. Symptoms include quality-related disorders in areas of social interaction, verbal and non-verbal communication, the variability of behaviours and specific learning disabilities. Abnormalities in development are already apparent in early childhood. **Aim:** To identify evidence for the clinical benefits of manual therapy of the musculoskeletal system in children diagnosed with ASD. **Methods:** The following databases and search interfaces were searched from Database start up until October 2015: Bio Med Central, Chiropractic Library Collaboration, Clinical Trials, Cochrane library, Dimdi, EBSCO host, Pubmed, Pubmed central, Medline Plus, Osteopathic research Digital repository, Osteopathic Research Web, and Physiotherapy Evidence Database. Further searches included journals provided by the University of Wales and the University of Duisburg-Essen. Studies were included if participants were children and young adults aged 0-21 years; studies published in English, German, or French; a diagnosis of autism or ASD, and study designs of randomized clinical trial, case-control studies, case series, case reports, and single subject studies (N of 1), which include manual therapeutic interventions of the musculoskeletal system. Two authors independently screened the studies for inclusion criteria, extracted the data and assessed for risk of bias. Methodological quality of randomized clinical trials was assessed by the Downs and Black tool. Quality of reporting for case series and case reports was assessed with the appropriate checklists provided by the QUALity and Transparency Of health Research (EQUATOR) network. **Results:** Included in the review were one randomized clinical trial (uncontrolled), one case series, and 11 case reports. The methodological quality of the included randomised clinical trial was rated as being poor. Quality of reporting for the included case series and case reports was also insufficient. All included studies used spinal manipulation, and indicated an improvement in autistic symptoms after the manual therapeutic intervention. **Conclusion:** The results of this systematic review confirm a general lack of good quality, high level of evidence studies on the topic, as well as no existing experimental studies that have been published in the last 10 years. This review indicates that the literature on the effects of chiropractic interventions to the musculoskeletal system of autistic children and young adults appears to be favourable with respect to the severity of their symptoms. However, the results of this review have to be interpreted with great caution, as the majority of identified studies were case reports. Further feasibility and pilot research is needed to lay the foundation for good quality clinical trials of spinal manipulation in the autistic child population.

Introduction

Autism spectrum disorders (ASD) are defined as a range of conditions classified as neurodevelopmental disorders, including autistic disorder (autism), Asperger syndrome, and pervasive developmental disorder (PDD).¹ Symptoms are quality-related disorders in the areas of social interaction, verbal and non-verbal communication, and a variety of behaviours and learning disorders.¹ Abnormalities in development are apparent already in early childhood and have a high variability.^{1,2} ASD is reported to occur in all racial, ethnic and socioeconomic groups, with about 1 in 68 children

being identified with ASD in 2012 in the US.³ Boys seem to be 4.5 times more often affected than girls.³

With respect to the causation of autism, no universally accepted explanations are currently present. The possibility of a genetic component, environmental triggers and imbalances in the neurophysiological state are all discussed.^{4,6} No specific medication is available to address autism. Medication is generally used to treat accompanying symptoms and modify behaviors of the condition.⁷ Current clinical guidelines recommend a combination of behavioral, devel-

opmental and educational approaches in order to enhance communication, social and cognitive skills and at the same time minimising autistic symptomology.^{8,9} Medication is recommended in case of challenging behaviour and other accompanying symptoms that justify its use.⁹

Use of complementary and alternative treatments is common among children with autism, 50 to 75% of children with autism may be treated with complementary and alternative medicine (CAM). The use of manipulative or body based therapies, such as Craniosacral therapy, osteopathic and chiropractic manipulation were reported by approx 25% of children with ASD.^{10,11}

Significant improvements in the symptomology in children with autism are anecdotally reported by chiropractors and other manual therapists, however, the research literature on the effectiveness of manual therapies in ASD is scarce.¹² A previous systematic review on the use of chiropractic care in children with ASD identified five studies (one randomized clinical trial, one case series and three case reports).¹³

All identified studies reported on an improvement in symptoms related to autism after the chiropractic intervention, however, the authors highlighted several limitations of the study designs, particularly of the randomized clinical trial and the case series, and concluded that further research was warranted.¹³

A methodological quality assessment was not formerly conducted in this review, as is recommended in the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines for systematic reviews.¹⁴ The objective of this present systematic review is to provide an expansion and update of this previous systematic review, aiming to answer whether new research on the topic has emerged, and formerly assess the available evidence for methodological quality and risk of bias, as well as giving detailed recommendations for further research on the topic.

Methods

A systematic literature search was conducted from Database start up to October 2015, using the following databases and search interfaces: Bio Med Central (www.biomedcentral.com), Chiropractic Library Collaboration (<http://www.chiroindex.org/#results>), Clinical Trials (<http://clinicaltrials.gov>), Cochrane library (<http://www.thecochranelibrary.com/view/0/index.html>), Dimdi (<http://www.dimdi.de/static/de/db/index.htm>), EBSCO host (<http://www.ebscohost.com/>), Pubmed (www.ncbi.nlm.nih.gov/pubmed), Pubmed Central (<http://www.ncbi.nlm.nih.gov/pmc/>), Medline Plus (<https://medlineplus.gov/>), Osteopathic Research Digital Repository (<http://ostemed-dr.contentdm.oclc.org/>), Osteopathic Research Web (www.osteopathic-research.com), Physiotherapy Evidence Database (<http://www.pedro.org.au/>).

Further searches included journals provided by the University of Wales (<http://www.wales.ac.uk/en/OnlineLibrary/OnlineLibrary.aspx>) and the University of Duisburg-Essen (www.uni-due.de/ub). The investigated journals were: *Advanced Clinical Child Psychiatry, Journal of Autism and Developmental Disorders, British Journal of Chiropractic, Chiropractic and Osteopathy, Journal of Clinical Chiropractic Pediatrics, Complementary Therapies in Medicine, Journal of Complementary and Alternative Medicine, The Journal of Manipulative and Physiological Therapeutics, The Journal of the American Osteopathic Association, Osteopathic Medicine and Primary Care, Pediatrics, German Journal of Osteopathy, International Journal of Osteopathic Medicine, Journal Pediatric, Maternal & Family Health, Journal of Vertebral Subluxation Research, the Journal of Upper Cervical Chiropractic Research.*

The search strategy used was: (Chiropractic (MeSH) OR Manual Therapy OR osteopath* OR OMT OR Physiotherapy OR CAM OR Osteopathic Medicine (MeSH)) AND (Autism OR Autistic disorder (MeSH) OR Autistic spectrum disorder (MeSH)).

Inclusion criteria were children and young adults aged 0-21 years; studies published in peer-review journals in English, German, or French; a diagnosis of autism or ASD, and study designs of randomized clinical trials (either controlled or not), case-control studies, case series, case reports, and single subject studies (N of 1), which include manual therapeutic interventions of the musculoskeletal system. Exclusion criteria were other forms of pervasive developmental disorders, other study designs such as narrative/systematic reviews, and no manual therapeutic interventions of the musculoskeletal system.

Study selection and data extraction

Two review authors independently screened titles and abstracts of the articles identified through the database search. Full-text articles of potentially eligible studies were obtained and independently evaluated for inclusion. Disagreement between authors was resolved through discussion or by consulting a third review author. Data extraction was performed by two independent reviewers into a data extraction table. Any discrepancies were resolved by discussion.

Quality assessment of included studies

The methodological quality evaluation of randomized clinical trials was carried out by means of the modified Downs and Black checklist.^{15,16} It contains 27 questions that are rated with one point each. The modification relates primarily to the evaluation of the Power Item 14 with 1 point, instead of 0.5 points, as in the original version. This checklist has been evaluated both as a valid and reliable method and

represents a powerful tool for methodological evaluation of clinical studies.^{17,18} The quality is evaluated in four domains: reporting, external validity, internal validity/bias and internal validity/confounding. According to the final score of the assessment, the methodological quality of the study is categorised into strong, moderate, limited or poor.^{16,19}

For the evaluation of the methodological quality of single subject studies (N of 1), case reports and case series, no checklists are currently available which have been analysed with respect to their validity and reliability. In order to introduce some quality measure, the quality of the reporting of these studies has been assessed in this review instead. The Enhancing the QUALity and Transparency Of health Research (EQUATOR) network states that readers may not be able to make reliable decisions on how relevant this research is with regards to clinical practice, how well the study was conducted and how reliable its findings are without accurate and complete description of methodological aspects of a given study.¹⁴ Accurate reporting may also enhance the usability of research, and if incomplete, would undermine the value and usability of research.¹⁴ For the case series, the reporting quality tool of Carey and colleagues was used.²⁰ The authors identified characteristics that well-reported case series studies should address: defined study question; well-described study population; well-described intervention; use of validated outcome measures; appropriate statistical analyses; well-described results; discussion/conclusions supported by the data presented; and funding sources acknowledged.²⁰ For the case reports, the 13-item CAsE REport (CARE) guidelines of the EQUATOR network were utilized.¹⁴ For simplicity reasons, scores of 0-10 out of a total of 30 were rated as poor reporting, scores of 11-20 were rated as moderate, and scores above 20 were considered good reporting.

The reporting of this present systematic review is based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.¹⁴

Results

Database searches revealed a total of 2,232 citations, and 1,569 records were screened after duplicate removal, with 1,528 articles excluded (Figure 1, Table 1). Out of 41 articles which were evaluated by full-text assessment, 13 articles were selected for this review, including one randomized clinical trial, one case series, and 11 case reports, involving a total of 52 study participants/subjects (Table 2). Compared with the systematic review by Alcantara and colleagues,¹³ no new experimental studies have been published to date, although a series of new case reports are now available in the literature which have been published in the last five years.

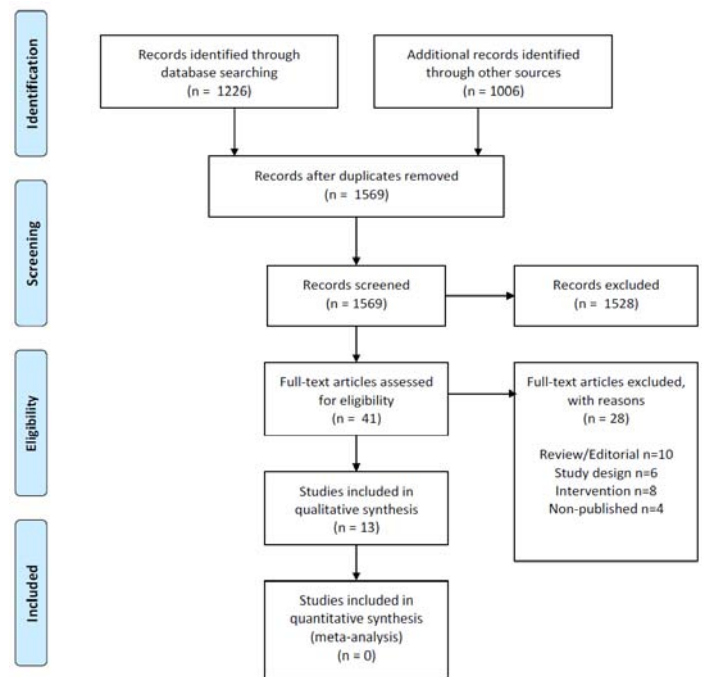


Figure 1: PRISMA Flow diagram of study selection.

Table 1. Database search results

Source	Number of studies retrieved
Clinical trials	3
Pubmed	154
Dimdi	569
Cochrane library	1
Pubmed Central	250
Medline Plus	28
EBSCO Host	79
Chiropractic Library Collaboration	33
Osteopathic research web	2
OstMed DR	3
BioMed Central	103
PEDro	1
Journals	1,006
Total	2,232

In all identified articles of this current review, chiropractic treatment was performed, with all of the studies describing manipulative techniques to the spine, either to the cervical or entire spine. Common to all studies is that positive effects, such as improvement in autistic symptoms, were observed in patients with ASD after treatment.

In all 11 case reports, chiropractic manipulation was performed one to three times per week, for up to 8 months (Ta-

Table 2. Overview of included articles

Study design	Number	Study
Randomised Comparison Trial	1	Khorshid (2006) ²¹
Case series	1	Aguilar (2000) ²²
Case report	11	Amalu (1998) ²³ Warner (1999) ²⁴ Neally (2003) ²⁵ Mc Cormick (2008) ²⁶ Hoffman (2008) ²⁷ Marini (2010) ²⁸ Cohn (2011) ²⁹ Cleave (2011) ³⁰ Scelfo (2011) ³¹ Noriega (2012) ³² Zielinski (2013) ³³
Total	13	

ble 3, next page). Age of patients ranged from 3-20 years. In two case reports, the cervical spine was treated and in the remaining nine other case reports, treatment of the entire spine was carried out. Thrust techniques were used in all studies (Table 3).

Likewise, the age range in the case series of 26 patients, conducted by Aguilar et al., ranged from 3 to 13 years, with similar frequency and duration of the treatment period (Table 3). Patients in this case series were treated by upper cervical spine chiropractic manipulation.

Participants in the randomized clinical trial, performed by Khorschid et al, were 4-16 years old, and treated in two groups twice each week for 3 months (Table 3). One group received upper cervical spine adjustment and the other group received spinal manipulation of the whole spine. The techniques commonly used in all studies were high velocity, low amplitude (HVLA) thrusts to correct segmental dysfunction; Thompson drop-assisted technique using a segmental 'drop table' for thrust enhancement; Activator method using an instrument to deliver a specific low force thrust; Atlas orthogonal adjustment using an instrument that transmits a mechanical impulse to the patient's atlas; and Torque release technique using a hand-held device for a torque and recoil-based adjustment. In the included studies, different assessment and outcome measures were used for the evaluation of data, both objective and subjective (Table 3).

In all studies, palpation was used to investigate the mobility of the spine, generally revealing improved flexibility of the spine by the end of the study (Table 3).

The five studies that used X-ray analysis show varying re-

sults.^{21,22,23,31,32} In the case report by Amalu, the subluxations of the spine were corrected after chiropractic intervention. Likewise, in the case report by Noriega, a decrease in subluxation of the atlas complex could be shown. Scelfo (2011), however, performed x-ray analysis only at the beginning of the treatment series and did not repeat it at the end, so no assessment of a change after spinal manipulation could be made (Table 3). In the study by Khorschid and colleagues, X-ray analysis was performed before and after the first cervical adjustment. Aguilar and colleagues performed X-ray analysis before the first treatment and prior to the fifth visit, when no subluxation was detected anymore, as assessed by leg length difference.

The leg length measurement in the prone position was applied in three of the studies assessed in this current review,^{21,22,32} resulting in the existing difference of leg lengths being corrected by the manipulative intervention.

The objective examination of patients with paraspinal thermography (thermal scan) was used in seven studies (Table 3). This diagnostic method is used to measure changes in temperature along the spine that may occur due to subluxations of spinal vertebrae. In the seven studies that applied the thermal scan, at the end of the investigation the spinal temperature was normalised and the vertebral subluxations were reduced or adjusted.^{23,24,26,27,29,32,33}

Surface electromyography (SEMG) was used in four studies as an evaluation tool for paraspinal muscle tension in vertebral subluxations; chiropractic treatments led to normalized values and thus to compensation of asymmetries (Table 3).^{24,26,27,29}

In four studies, the Autism Treatment Evaluation Checklist (ATEC) was used to assess the severity of autistic symptoms (Table 3).^{21,26,28,31} This checklist has been designed to monitor children with autism over time in order to evaluate potential effects of a given therapy.³⁴ This simple scoring scale with 77 items is filled in by parents or carers. It consists of four subdomains: speech/language, sociability, sensory and cognitive awareness, and health/physical/behaviour, providing a possible total score range of 0-180, with a higher score generally indicating a greater degree of impairment in symptoms. Total scores of less than 30 indicate that the child may lead a normal and independent life, scores of less than 50 indicate that the child may lead a semi-independent life, scores between 50 and 104 hint to moderate autistic symptoms, and with scores above 104 the child would be considered severely autistic.³⁴

Within the scope of this review, the clinical changes after chiropractic treatment were demonstrated by a decrease in the ATEC scores, with a reduction of the total scores from

Table 3. Overview of included studies

Study	N	Male: female	Study design	Age	Treatment approach	Techniques used	Outcome measures/diagnostic test applied	Results
Khorshid (2006)	14	13 : 1	Randomized clinical trial	4-16 yrs	Chiropractic treatment: Group 1: upper cervical spine; Group 2: entire spine, twice each week for 3 months	Instrument-assisted atlas orthogonal adjustment of cervical spine, no other techniques/ approaches described	ATEC; X-rays; spinal palpation; leg length difference; palpation in motion	Manipulation of the upper cervical spine led to reduction of ATEC scores by 32% in group 1 compared to 19% in group 2; in both groups, X-rays were normal, free movement of spine, leg length difference was resolved
Aguilar (2000)	26	21:5	Case series	3-13 yrs	Chiropractic treatment of upper cervical spine for 9 months 3 times per week	Orthospinology specific upper cervical technique	X-rays, leg length difference, CARS, modified autism rating scale	Statistically significant reduction of CARS scores after 9 months by 10%; statistically significant reduction of modified autism rating scores after 9 months by 22%; leg length difference resolved
Amalu (1998)	1	0:1	CR	5 yrs	Chiropractic treatment of upper cervical spine for 12 weeks	Thrust techniques for correction of Occipito-atlanto region	Thermalscan upper cervical spine; X-rays upper cervical spine; palpation in motion	Neurophysiological symptoms and temperature of the spine normalized, subluxations of spine resolved, follow-up after 8 and 11 weeks confirmed results; Diagnosis of autism reversed
Warner (1999)	1	0:1	CR	5 yrs	Chiropractic treatment of the spine for 3x per week, after 2 years 1x per week	Thrust techniques	SEMG; thermal scan; Palpation of spine in motion	Autistic symptoms positively influenced; reduction of vertebral subluxation; normalised thermal scan and SEMG
Neally (2003)	1	0:1	CR	19 rs	Six chiropractic treatments of the spine	Occiput: Drop-headpiece; thrust techniques to spine	Palpation of spine in motion	Reduction of autistic behavioural symptoms, improved spinal movement, patient started to speak
McCormick (2008)	1	1:0	CR	4 yrs	Chiropractic treatment of spine for 6 months	Thompson-Drop-Assisted Technique for S2; Thrust C1; Activator for Thoracic spine	ATEC; thermal scan; SEMG; palpation in motion	Reduction of autistic behavioural symptoms; social behaviour, language and understanding of environment improved; ATEC reduced from 97 to 90; normalised movement, temperature and EMG of spine
Hoffman (2008)	1	0:1	CR	3.5 yrs	Chiropractic treatment of the spine for 10 weeks	Torque Release Technique	SEMG; thermal scan; palpation in motion	Improved symmetry and balance of the nervous system after treatment series and follow-up after 2 months; normalised EMG and thermal scan; subluxations of spine resolved; subjective and objective change in behaviour, as reported by parents
Marini (2010)	1	1:0	CR	6 yrs	Chiropractic treatment of spine and pelvis for 16 weeks	Thompson-Drop-Assisted Technique for SIJ; thrust techniques for thoracic and lumbar spine	ATEC; palpation in motion	Reduction of ATEC scores in: language from 16 out of 28 to 9 out of 28; social behaviour from 16 out of 40 to 7 out of 40; perception from 15 out of 36 to 6 out of 36; health from 23 out of 75 to 10/ out of 75; total from 70 out of 180 to 32 out of 180 (improvement of 54,3%); improvement of symmetries of spine and pelvis
Cohn (2011)	1	1:0	CR	3 yrs	Chiropractic treatment once per week for 10 weeks	Correction of spine, not described in detail	SEMG; thermal scan; palpation in motion	Muscle tonus and posture, reading, stereotypes and social behaviour positively influenced; language development positively influenced; SEMG and thermal scan normalized reconstitution of spinal symmetry
Cleave	2	1:1	mCR	17 yrs (female) and 20 yrs (male)	20 chiropractic treatments of spine and pelvis for 5 months	Patient 1 (male): Activator method chiropractic technique Patient 2 (female): Thompson-Drop-Assisted Technique, AMCT	Palpation in motion	Improvement of social behaviour; ability to concentrate; reduction of aggressive behaviour; increase of tolerance to frustration and perception, reported by caregivers improvement of symmetry of spine and pelvis

Study	N	Male: female	Study design	Age	Treatment approach	Techniques used	Outcome measures/diagnostic test applied	Results
Scelfo (2011)	1	1:0	CR	9 yrs	16 Chiropractic treatments 2x per week	Thrust and later toggle-recoil for Atlas; Thrust SIJ	ATEC; palpation in motion; x-rays	Improved ATEC scores in: language from 16/28 to 14/28; behaviour from 20/40 to 15/40; perception from 18/36 to 15/36; health from 42/70 to 22/70 and total from 96/180 to 66/180; reconstitution of spinal symmetry; reduction of medication use, no X-ray assessment at end of study
Noriega (2012)	1	1:0	CR	6 yrs	Chiropractic treatment of upper cervical spine 2x per week for 12 weeks, additional 3 weeks 1x per week	Thrust (triceps pull)	Static Palpation; palpation in motion; postural pelvis-evaluation; weight distribution; leg length difference; thermal scan; X-rays	No neurological and musculoskeletal findings reported; leg length difference restored; symmetric thermal scan, reduction of subluxation of atlas complex; resolution of nocturnal enuresis; improvement of ASD by 70% particularly in areas of school performance and social behaviour
Zielinski (2013)	1	0:1	CR	3 yrs	Chiropractic treatment of cervical and thoracic spine 1x per week for 8 months	Treatment 1-11: Activator-method; from 12th treatment: manual thrust techniques	Palpation in motion; postural evaluation; thermal scan	Subluxations decreased; thermal scan more balanced; vegetative imbalance reduced; ASD-symptoms improved in area of behaviour; child started to speak; eye contact has been established; total resolution of bilateral headache, reflux, nausea and insomnia; no epileptic fits; use of medication terminated

Abbrev.: ASD — Autism Spectrum Disorders; AMCT - Activator Method Chiropractic Technique; ATEC - Autism Treatment Evaluation Checklist; CARS — Childhood Autism Rating Scale, CR — Case Report; mCR — multiple case report; RCT — Randomised Comparison Trial; SEMG — Surface Electromyography; SIJ — Sacro-iliac joint

97 to 90 out of 180,²⁶ from 70 to 32 out of 180 (corresponding to a reduction by 54.3%),²⁸ and from 96 to 66 out of 180.³¹ In the study by Khorschid et al., mean ATEC values in the full spine adjustment Group changed from 68 to 46 out of 180 (calculated by the authors of this current review, as only individual values of participants were given in the original publication), and the mean values of the cervical spine adjustment Group changed from 41 to 30 out of 180.²¹ In regard to the safety of the spinal manipulative treatment approach, no adverse events were reported in the studies. The assessment of the methodological quality of the ran-

domized clinical trial revealed 6 out of 27 points, corresponding to an overall poor methodological quality. In all domains (reporting, internal and external validity), serious shortcomings were detected, including selective recruitment of the study population and an absence of a sample size calculation, lack of blinding, no statistical analysis of data rather than individual patient data being presented in tables. Neither probability values (p-values) nor confidence intervals were calculated or reported (Table 4).

The reporting quality of the included case series achieved

Reporting									
1	2	3	4	5	6	7	8	9	10
N/U	Y	N/U	Y	N/U	Y	N/U	N/U	N/U	N/U
External Validity									
11	12	13	14						
N/U	N/U	Y	N/U						
Internal Validity/Bias									
15	16	17	18	19	20	21			
N/U	N/U	N/U	N/U	N/U	N/U	Y			
Internal Validity/Confounding									
22	23	24	25	26	27				
Y	N/U	N/U	N/U	N/U	N/U				
Total Score = 6/27									
Methodological Quality: Poor									
*Modified Downs and Black checklist ^{15,16} ; Abbrev.: Y — Yes, N/U — No/Unclear									

a poor three points out of a possible eight points, with a clearly defined study question, study population and intervention as well as appropriate statistics missing (Table 5).

Reporting of all 11 case reports was assessed using the 13-item (including several subitems) CARE checklist. Out

of a total possible 30 points, one study achieved under 10 points, corresponding to overall poor reporting (Table 6). Seven case reports achieved scores between 10 and 20, corresponding to overall moderate reporting. Three case reports achieved scores of 20 and higher, corresponding to good reporting.

Discussion

This systematic review update included in total one randomized clinical trial, one case series, and 11 case reports. Since the publication of the randomized clinical trial by Khorschid and colleagues in 2006, no new experimental evidence has been generated/published in the last 10 years.

Reasons for this can only be speculated on, which may form part of resource implications of chiropractic research, i.e. a lack of highly qualified and experienced research staff and research funding, a lack of public interest on the topic, or a

Table 5: Reporting quality of case series by Aguilar et al (2000)

Study characteristic	Presence
Study Question	No
Study population	No
Well-described Intervention	No
Valid outcome measure	Yes
Appropriate statistical analysis	No
Well-described results	Yes
Discussion/Conclusion supported by data	No
Funding source acknowledged	Yes

SOURCE: (Carey 2003) 20

Table 6: Reporting quality of case reports

	Amalu 1998	Warner 1999	Neally 2003	McCormick 2008	Hoffman 2008	Marini 2010	Cohn 2011	Scelfo 2011	Cleave 2011	Noriega 2012	Zielinski 2013
1	N	Y	N	N	N	N	N	N	N	N	N
2	N	N	N	Y	N	Y	Y	Y	Y	Y	Y
3a	N	N	N	N	N	N	N	N	N	N	Y
3b	N	Y	N	Y	Y	Y	Y	Y	Y	Y	Y
3c	N	Y	N	Y	Y	Y	Y	Y	Y	Y	Y
3d	N	N	N	Y	Y	Y	Y	Y	Y	Y	Y
4	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
5a	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
5b	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
5c	Y	N	N	Y	Y	Y	N	Y	N	Y	Y
5d	Y	Y	N	Y	Y	Y	Y	Y	N	Y	Y
6	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
7	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
8a	Y	Y	N	N	Y	Y	N	N	N	Y	Y
8b	N	N	N	N	N	N	N	N	N	N	N
8c	N	N	N	N	N	N	N	N	N	N	Y
8d	N	N	N	N	N	N	N	N	N	N	N
9a	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
9b	Y	Y	N	Y	Y	Y	Y	Y	Y	Y	Y
9c	Y	Y	N	N	N	Y	Y	Y	Y	Y	Y
10a	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
10b	N	N	N	N	N	Y	N	N	N	Y	Y
10c	N	N	N	N	N	N	N	N	N	N	N
10d	N	N	N	N	N	N	N	N	N	N	N
11a	N	N	N	N	N	Y	N	N	N	N	Y
11b	Y	N	N	Y	Y	Y	N	Y	Y	Y	Y
11c	N	N	N	N	Y	Y	N	N	Y	Y	Y
11d	N	Y	N	Y	Y	Y	Y	Y	Y	Y	Y
12	N	N	N	N	N	N	N	N	N	N	N
13	N	N	N	N	N	N	N	N	N	N	N
Total 13	15	7	16	17	17	21	15	17	16	20	23

SOURCE: (Gagnier 2013) 14,54, Abbrev.: Y – Yes, N – No

misinterpretation of the available evidence in itself.

Effects of chiropractic treatment on symptoms

Common to all studies/reports within the scope of this review is that positive effects such as improvements of autistic symptoms have been observed in patients with ASD after treatment. However, due to a lack of statistical data and poor study design of the included randomized clinical trial, no comments on the effectiveness/efficacy of the chiropractic intervention can be made.

In four studies of this review, the ATEC was used to assess the severity of autistic symptoms. According to the Autism Research Institute, ATEC is a validated and reliable instrument, available free of charge for research purposes.³⁴ This questionnaire is particularly suitable for the detection of improvement or decline of autistic symptoms, provided the same person fills out the questionnaire.³⁴ The advantage of the ATEC is the extent to which a treatment leads to changes in the expression of autism in the assessment. Other checklists, such as the Childhood Autism Rating Scale (CARS), the Gilliam Autism Rating Scale (GARS) or the Autism Behaviour Checklist (ABC) can be used to establish the diagnosis of autism, but do not judge the benefits of having a therapy for an autistic child.³⁵ ATEC is an ideal measuring tool for the collection of reliable data in the context of interventional studies.^{34,35}

It was noted, that in the randomized clinical trial by Khorschid and colleagues, children with different severities of autism were participating, based on ATEC scale assessments. Due to the very small sample size of 14 patients in this trial, random group allocation of these patients was not uniform, resulting in 2 and 1 severely impaired patients (scores >104), 2 and 0 moderately affected patients (scores >50), 1 and 2 patients with scores >30 and 1 and 4 patients with very mild symptoms (scores <30) in the groups of full spine adjustment vs cervical spine adjustment, respectively. These imbalances in symptom severity are to be avoided in future clinical trials in order to avoid skew of data. This can be achieved by strictly defining the inclusion and exclusion criteria, taking into account the practicalities of patient recruitment for clinical studies in real-world settings. Severely autistic children may not be easily accessible, and ethical considerations in the future study design must be taken into account, such as supply of standard care for all patients in the study, regardless of group allocation. Since no other randomized clinical trial exist other than the study by Khorschid et al, it is difficult to predict which patient group with a given symptom severity may benefit most from chiropractic treatment, if there is an effect after all.

Analysis of a considerable number of single case studies on autistic children may be the way forward in establishing

which patient group may be most suitable for engagement in future clinical trials, which techniques to use and which areas of the spine to treat. Single subject studies (N of 1) are ideal for exploring treatment conditions for individual patients, and where heterogeneity of the patients may limit generalisability from trial data.^{36,37}

On practical grounds, for example a group of chiropractors who regularly treat children with autism could be engaged by performing simple single subject studies (N of 1), whereby each parent of a presenting autistic child is asked to fill in the ATEC online before each treatment, so that a potential change in symptom severity can be closely monitored. Additionally, participating chiropractors should report on techniques used and body areas treated.

The case series by Aguilar and colleagues report on the changes of each individual child after chiropractic intervention, and in several cases parents reported a decrease of medication use and improvement on chronic infections.²² Since these aspects are not monitored within the ATEC questionnaire, they could be assessed and reported as part of the case note taking prior to treatment, and reassessed before each subsequent follow-up treatment, with these data also feeding into future study results.

The randomized clinical trial in this current review did not incorporate a follow-up assessment after the end of the treatment period, hence the duration of treatment effects, if any, are not clear. Future studies could address this question, again by initially performing single subject studies (N of 1) in order to explore the number of treatments needed and time for potential treatment effects to last. Additionally, potential confounders and the feasibility of study procedures, such as number and time required and adherence to fill out questionnaires, as well as attendance at study appointments can be explored. Since outcome measures with respect to autism symptoms will mainly be based on questionnaires and medication use, the acquisition of robust baseline data during 3-5 weeks before the first chiropractic intervention (run-in phase) may be indicated, in order to assess the variance in change of these data during day-to-day life of the participants.

Influence of confounders

One common problem in clinical studies is the reporting of confounding factors which have the potential to influence study results. According to Skelly, confounders commonly may indicate an association between a treatment modality and an outcome, when in fact, there is no real association.³⁸ Confounding variables such as age, sex, diet, exercise, natural course of disease, lifestyle changes, emotional events, comorbidities etc should be considered in the design and reporting of clinical studies. Skelly recommends adequate

reporting of patient characteristics/demographics as well as definition of inclusion criteria by specific confounding variables.³⁸ However, it is noted that there always will be residual confounding, no matter how many variables one accounts and adjusts for, most likely due to unknown factors.³⁸

In the included studies in this current review, no attention to confounding factors was paid. In order to address and explore this issue for future clinical trials, parents and carers may be most suitable to answer questions with respect to effects of diet changes, emotional events, lifestyle changes such as holidays etc on the symptoms of their autistic child. They also may be able to report on patterns of symptom changes which may be related to season of the year or specific weather conditions, or change of symptoms over the last few years. These questions can be best explored as part of a qualitative interview study where parents/carers have the opportunity to express their feelings/attitudes/beliefs about the subject of discussion, but findings of these qualitative studies should be verified by larger scale surveys of parents with autistic children, in order to get a more realistic picture of the population to be studied.

The ethics of effective treatment supply

In the randomized clinical trial of Khorshid and colleagues, two different treatment approaches were compared. It is not clear, whether the participating children received other forms of treatment, such as usual care. Since chiropractic care in autism cannot be considered effective to date due to lack of evidence, other existing effective treatments, if applicable, should be provided for future study participants. In order to meet the rigorous ethical requirements for the conduct of clinical trials involving children, the current clinical guidelines on the treatment of children and young adults with autism should be taken into account in any future study design, in that all study participants should receive the recommended therapies, and additionally the chiropractic manipulation as the specific intervention to be explored. Importantly, this would avoid the deprivation of effective care of all study participants, independent of group allocation, but also would likely encourage interdisciplinary communication and increase acceptance for such studies within the medical profession.

Safety aspects

Assessment of adverse events as part of the chiropractic treatment was not mentioned in the identified studies. This could be interpreted as no adverse events were taking place, or that adverse events occurred but have not been reported. Incidences of serious neurological and/or vertebrobasilar complications in all patient groups were estimated as one in 250 million chiropractic visits³⁹ which was challenged by other authors, stating that they feel it was inaccurate and

likely to be underestimating risk.⁴⁰ Survey data from chiropractic practices indicate that one child per 100-200 attending may have a mild adverse event not lasting longer than 24 hours, with no serious adverse events reported.⁴¹⁻⁴³ Since the literature on the safety of spinal manipulation as part of experimental studies in children is scarce and controversial, it is vital that, regardless of study design, adverse events are assessed and reported on; even if no adverse events occurred, this should be equally stated.

Methodological quality and reporting

The assessment of the methodological quality of the randomized clinical trial in this review was performed by means of the Downs and Black assessment tool which is valid and reliable and can serve as a strong methodological instrument for the evaluation of methodological quality.¹⁵⁻¹⁸ The weakness of this instrument, however, is the reliability in the field of external validity, which is rated as low.¹⁵ The advantage of this checklist is that it can be used for the evaluation of randomized and non-randomized trials in health research¹⁵ and currently is in this respect one of the best assessment tools for studies involving public health interventions.¹⁸

Although the RCT by Khorshid and colleagues is rated by design on top of the hierarchy of evidence,⁴⁴ the methodological quality evaluation resulted in only 6 out of 27 points, achieving a poor quality rating. Furthermore, it only achieved a low scoring in the categories of internal and external validity. No statistical methods were applied so that the effects of chiropractic treatments of the upper cervical spine compared with those of the whole spine in autistic children are unclear. Since this trial compared two different treatments with each other, blinding of subjects with respect to a 'real' intervention and a sham procedure was perhaps not anticipated by the the study authors. Nevertheless, future clinical trials should make an attempt to control for non-specific effects, by taking an appropriate sham control as well as a 'no intervention' control into account, all in addition to usual care, as only then one can conclude on the effectiveness of the chiropractic intervention and a potential placebo effect of non-specific elements of such a treatment session, such as patient expectations, practitioner-patient relationships, practice environment, and the manual touch itself.⁴⁵ Appropriate sham controls in clinical trials of spinal manipulation have been described in previous studies.⁴⁶⁻⁵³

Future studies with randomized clinical trial design should adhere to the CONSORT reporting guideline as specified by the EQUATOR network.¹⁴ Likewise, in order to improve the quality of reporting for future single subject studies (N of 1) and case reports, the appropriate guidelines should also be considered and adhered to.¹⁴ In the current review, the reporting of the case reports in general lacked information

on diagnostic challenges and reasoning, prognostic features of the condition and a shared patient perspective as well as informed consent. Arguably, these aspects may not be seen as being relevant in the day-to-day practice encounter of autistic children and young adults or are performed as part of the routine assessment and treatment anyway, however, they should be reported on in order to meet the rigorous standards of reporting to guide clinical practice.⁵⁴

Future directions

This review highlights a scarcity and poor methodology of clinical trial data. In order to move forward and establish a solid base for future research, single steps in the form of initial pilot work need to be taken. Several authors have emphasized a focus on treatment outcomes research being best conducted in phases.⁵⁵⁻⁵⁷ Phase 1 would be the exploration of a new treatment with a small number of individual patients to test the therapeutic effect. As previously mentioned, this could be achieved by performing single subject studies (N of 1). After optimization through further studies and determining the optimal treatment candidates, the potential efficacy of the treatment should further be explored in small pilot studies (Phase 2). The third phase would then be well-controlled trials that test the efficacy under ideal conditions, and if efficacy has been shown in this phase, the treatment then should be tested within the real-world setting and a cost-effectiveness analysis would be added (Phase 4 and 5).⁵⁵⁻⁵⁷

Review limitations

This present systematic review has the following limitations which may have influenced the results presented: Only English, German and French language articles were included, which may have led to the exclusion of relevant articles in other languages. Authors of original publications were not contacted for additional information on data, and database searches were restricted to peer-reviewed literature only. Additionally, inclusion of so-called 'grey literature' may reduce potential publication bias in future systematic reviews on the topic.

Conclusion

The results of this systematic review update confirm a previously reported general lack of good quality, high level of evidence studies on the topic, as well as no existing experimental studies that have been published in the last 10 years. The authors' interpretation of the results of this review indicate that chiropractic intervention to the musculoskeletal system of autistic children and young adults may have the potential to create a positive effect on the severity of their symptoms. However, the majority of study designs identified were case reports/series with only individual patients studied and reported on, hence generalisation of results to the autistic child/youth population are not indicated.

Therefore, the results of this study have to be interpreted with caution, despite positive results being presented with regards to autistic symptoms. Further feasibility and pilot research is needed in order to formulate a robust study hypothesis and design a high quality randomized controlled trial to explore the effectiveness and efficacy of spinal manipulation in the autistic child and young adult population.

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Development and testing of a multidimensional parent reported outcome measure for common presenting complaints of infancy: the UK infant questionnaire.

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ABSTRACT

Objectives: Patient reported outcome measures are recognized as important and valuable tools to monitor patient progress in healthcare. It is fundamental to clinical practice to understand whether the treated patient has improved or not. Despite the highest use of outpatient healthcare among all pediatric age groups, no age-appropriate outcome measures are available for the infant. Therefore, the objective of this study was to develop and test a new infant outcomes instrument for the most common presenting complaints of infancy. **Methods:** This was a multi-phase study designed to develop a questionnaire using maternal interviews and to test it for reliability and validity for use in well child clinical practice. After collecting the mother's views, grounded theory and content analysis were used to derive themes and domains for the questionnaire. After achieving face validity, the instrument was evaluated for test-retest reliability, homogeneity and concurrent criterion validity. Subjects comprised a convenience sample of mothers who presented their infants to a university-affiliated chiropractic teaching clinic on the south coast of England. **Results:** Maternal interviews revealed mothers' concerns about feeding, sleeping, crying and other aspects of infant activities of daily living resulting in construction of a 12 question instrument. The questionnaire showed excellent test-retest reliability (ICC = 0.96) and good internal consistency (Cronbach's α = 0.8). In validity testing, ten questions showed positive correlation to a statistically significant degree against their established gold standard references. In all, 294 mother/infant dyads were involved in the research project. **Conclusion:** The UK Infant Questionnaire is the first parent reported outcome measure for use with the most common complaints of the infant patient based on maternal views. As such, this instrument meets the standard set by the UK National Health Service to involve the parent's voice in their child's care, and is therefore innovative in its field. Although further testing is indicated, and we make no claims that this instrument is comprehensive in all aspects of infant well-child care, it may be used by individual clinicians in routine daily practice to gain understanding of clinical progress of individual patients.

Keywords: Outcome measures, Pediatrics, Chiropractic

Introduction

The infant (0-1 year of age) incurs the highest health care costs of any age throughout childhood.¹ Common complaints are crying, feeding and sleeping problems, which are reported in 20-33% of infants,^{2,3} and an additional 15% of infants are afflicted with two or more of these issues.³ Problems in early infancy are associated with short term risks, including early discontinuation of breastfeeding,^{4,6} infant abuse,^{7,8} maternal depression,^{8,9} and long term risks including developmental problems.³ As birth injury has been largely implicated in these early infant complaints,¹⁰ biomechanical factors may have some degree of influence on the short and long-term prognoses of these conditions.¹¹

Chiropractic care is a modality commonly sought by fami-

lies, and large numbers of infants are seen by chiropractors.¹²⁻¹⁴ Despite high usage of chiropractic care for infants, the profession has been criticized for a lack of evidence upon which to support pediatric chiropractic care.¹⁵ However, virtually all branches of health care have been guilty of too little evidence-based-practice for children.^{16,17} A crucial example in medicine is the alarming and continued use of off-label prescriptions, in the pediatric population, whilst recognizing the lack of evidence for safety and efficacy required by regulatory standards.^{16,17} A key reason for the scarcity of high quality research in pediatric care, across all health care arenas, is the paucity of relevant outcome measures in routine practice for this age group.¹⁸ The development of age-appropriate outcome measurements is a pragmatic and appropriate next step considering the high

usage of health care in the first year of life.

Background

Patient reported outcome measures (PROMS) have been increasingly utilized by healthcare communities to measure patient responses to treatment. Their use in research is well known, as PROMS were initially developed to enable a clinician to measure outcomes in clinical trials and to take into account a patient's subjective health status and quality of life.¹⁸ The routine use of outcome measures is increasingly called for in all types of health care as a valuable measure of change, be it improvement or worsening, in order to document whether or not a specific treatment has value to the individual patient. Collecting outcome data from large numbers of patients in chiropractic practice has been proven feasible.¹⁹

Including children in the endeavor to use patient choices to improve their care is crucial.^{20,21} Pragmatic and efficient outcome instruments for the infant patient would be particularly useful because other methods to study responses to care are onerously time and cost consuming. For example, large scale direct observation of the infant with video-recording or in-house recordings would be invasive and problematic for both the family and the researcher. Diaries have been used as the gold standard (validated against in-house recordings) for the infant's behaviour,^{22,23} but these are time consuming for both parents and researchers and are therefore, under-utilized, ignored or abandoned. Questionnaires may be a practical replacement for diaries as an efficient tool to measure outcomes, provided they can be documented as equally credible. Mothers have been shown to be reliable reporters of their infant's behavior,²²⁻²⁴ and therefore intake and discharge questionnaires could be a quick, accurate and pragmatic way to study this population's behaviors and treatment results.

A small number of questionnaires have been developed for individual aspects of the infant's problematic behavior, including sleeping,²⁴⁻²⁶ pain,^{27,28} crying,²⁹ and feeding.³⁰ Despite these focused attempts, there is no established validated pediatric instrument that covers all key aspects of an infant's behavior.²⁴⁻³¹ Many individual problems faced by the infant population (feeding, crying, sleeping, postural problems/pain) are overlapping and interlinked,² and as such should be viewed as a piece of the clinical puzzle, rather than the whole picture.

Because of the requirement to represent the exact needs of the patient,¹⁸ with infant patients the maternal voice must be heard by the researcher. Qualitative approaches used to ascertain what is most important in infant health care from the mother's perspective can be considered the key foundation for any new outcome measure.^{32,33} This not only

ensures content validity, but focuses on and respects the voices of those for whom the outcome measurement is intended, which in this case is the mother of the infant.

Therefore, the goals of this project were to ask mothers what key concepts were most important in their infant's health, use these domains to develop a questionnaire, investigate the reliability and validity of the questions in the instrument and test it for intake and follow-up to understand parent report of outcomes of infant care.

Methods

The development stages and exploration of instrument testing are shown in Table 1 (next page) which was generated to summarize the methodological procedures to be viewed at a glance.

Subjects and setting: A convenience sample of mothers who presented their infant to a chiropractic teaching clinic was recruited. Inclusion criteria were English fluency and consent to be part of the study. The treating clinician was not aware as to whether the mother had enrolled in the study or not.

Ethical approval was granted by the AECC Research Ethics Subcommittee in July 2014. There was no funding for this project and no financial incentive for either the subjects or the investigators.

Phase 1: Qualitative Study: Mothers attending the clinic with their infant were asked to take part in an interview. Interviews were based on a discussion guide,³² broadly based on common presenting complaints in the infant age group and outcomes and experiences of healthcare. This document was evolving and was changed as new topics arose in interviews, a principle used in reflexivity.^{34,35} Interviews were anonymously audio-recorded and transcribed verbatim.

An exceptionally large sample size in the qualitative phase was determined for the purpose of obtaining as many opinions from the mothers as was reasonably possible. Data saturation is generally considered complete after 12 interviews.³⁶ Brod et al. suggested that after 12 interviews, between 88% and 92% of analysis codes (themes or domains) could be identified.³⁶ However in this study it was decided to continue qualitative research until no new themes had emerged for several interviews in order to be assured of saturation. This was important to ensure high content validity of the domains for use in the questionnaire.

Concepts from content analysis and grounded theory were used to extract data from transcripts, based on work by Lasch et al. and Brédart et al.^{32,33} Themes were the topics

which featured most heavily throughout each transcript, and were decided after each researcher individually read each transcript, and discussed to reach agreement by at least three researchers.

Phase 2: Development of the questionnaire: Following data analysis, each team member wrote questions around the themes and domains from the interviews. Questions were subsequently reworded to utilize the mothers' language in order to be conscious of the subjects' needs and parlance.³²

Phase 3: Face validity: Because this was a new type of instrument, never before tested, it was considered that face validity should be established to determine whether the questionnaire has merit or "face value." Experts in the fields of either pediatric chiropractic or research were asked for feedback on all aspects of the questionnaire including topics covered, anchors for the answers to each question, and the wording and layout. An 11-point scale was used with descriptive anchors at zero, five and ten, which varied depending on the question. Feedback was recorded and dis-

cussed with the team before implementing changes.

As the central focus of this project, mothers were given the final consideration and asked both to complete and comment on two versions of the questionnaire. A convenience sample of mothers who presented their infant to the clinic for care were observed during the completion for any hesitation and the time taken to complete. They were asked what each question meant to them and if they had any issues with filling out the questionnaire and if any key points were missing. The questionnaires were then re-formatted and the final version of the United Kingdom Infant Questionnaire (UKIQ) was generated ready for reliability and validity testing.

Phase 4: Reliability: Test-retest reliability: First, test-retest reliability was measured by asking mothers to complete the same questionnaire twice on the same day. The items on the second questionnaire were given in a different order to reduce the possibility of the participant memorizing their initial response,^{38,39} although it was considered that this risk

Table 1. Summary of Methods to Develop and Test an Infant Outcomes Instrument

Inclusion criteria: Mothers of infants <12 months, fluent in English

Exclusion criteria: Mothers of infants >12 months, not fluent in English

Subjects and setting: Convenience sample of mothers presenting to the AECC outpatient clinic Ethics Research proposal approved by AECC Research Ethics Subcommittee

• **Phase 1: Qualitative Study**

Interviews and focus groups were conducted using a discussion guide. Topics included experiences of care and aspects of healthcare important to the mother. Data analysis: Grounded theory content analysis was used to create themes.

• **Phase 2: Development of questionnaire**

Themes from interviews were used to create questions corroborated with research into infant public health issues. Questions were generated to reflect maternal concerns and worded to reflect maternal language.

• **Phase 3: Face validity**

Experts

Experts in either pediatric care or research were asked to give feedback on the questionnaire during interviews or focus groups.

Mothers

Two questionnaires (versions one and two) with the same layout but different wording were completed by mothers. The time needed to complete the questionnaire, signs of hesitation, erasures or skipped questions were observed. Feedback from mothers was recorded and integrated.

• **Phase 4: Reliability**

Test-retest reliability

The questionnaire was given to mothers twice on the same day to determine test-retest reliability. The questions on the retest questionnaire were reorganized to avoid memory aids.

Homogeneity (internal consistency)

Internal consistency was calculated over each of the administrations of three questionnaires (including pre- and post-treatment) using Cronbach's α and item corrected total correlations.

• **Phase 5: Validation of the questionnaire**

Responsiveness

Mothers completed a questionnaire pre- and post-treatment.

Six-day behaviour diary

Mothers completed a 24-hour behaviour diary (gold standard) for six consecutive days. After six days the diaries were returned to the researcher and mothers completed the UKIQ and its correlated validation questionnaire.

Validation

Each question of the UKIQ was correlated to a question out of a reference questionnaire or the six day behaviour diary using Spearman's ρ , Phi Coefficient, Cramer's Phi or Cohen's Kappa depending on data requirements.

• **Phase 6: Pilot testing**

Questionnaire was implemented in a busy infant practice to test the practicalities of usage from clinician, parent and office staff perspectives.

was low with fatigued and stressed new mothers completing the forms. The test-retest reliability was calculated using intra-class correlation (ICC) coefficient two-way mixed single measures (ICC3.1).³⁹⁻⁴² It assessed the reliability of ratings by comparing the variability of different ratings of the same subject to the total variation across all ratings and all subjects.

Homogeneity: Second, homogeneity (internal consistency) was assessed. Homogeneity measures whether all of the items in the questionnaire are tapping different aspects of the same attribute. If this is the case, the items in the questionnaire can be added to give a total score.³⁸⁻⁴² Cronbach's α statistic⁴⁰⁻⁴⁴ and item-corrected total correlations (Pearson's correlation coefficient, r)⁴⁰ were used. Cronbach's α provided a measure of the internal consistency of the questionnaire in its totality. Cronbach's α uses inter-item correlations to determine whether constituent items are measuring the same domain;^{44,45} it compares the variance of each question with the variance of the total score. Item corrected-total correlation is calculated on a per-question basis and shows whether individual items are correlated to the total score of the questionnaire.

Phase 5: Validation of the questionnaire: Although many widely used instruments in health services have never been tested for validity,⁴² it was decided that reliability alone was insufficient to support the use of this instrument. Validity was tested to determine the degree to which the instrument measured the domains it purported to measure, by testing each question against its own validated measure. Validity is measured by degrees and is not a binary judgement. Validity testing was carried out in stages to test face, construct and criterion concurrent validity.

Validity: Each domain of the questionnaire was matched to an external measure that was the gold standard for that dimension (Table 2, next page). This criterion-related approach seeks the amount of correlation with another test designed to measure the same thing. Because the UKIQ was the first questionnaire developed directly from the current views of mothers, some of the topics were novel and had not previously been investigated, only a reference standard of a six day 24-hour behaviour diary could be used, as diaries have been validated as accurate records of infant behavior.²²⁻²⁴ The gold standard questions were then compiled to form a reference questionnaire. This was required because it was considered unethical to ask mothers to complete eight questionnaires along with a diary for comparison at one time.

Mothers were given the UKIQ, the six-day diary and the reference standard questionnaire at indicated times during the infant's intake, treatment and follow-up. Data from each

survey were entered into SPSS and statistical tests were chosen relative to the type of data tested.

Analysis: Because of the nature of the data, non-parametric statistics were predominantly used. Each question was tested more than once, if more than one appropriate test could be used. Pearson's r tested scale based data. The phi coefficient was used for nominal based questions (yes/no). Where questions had more than two categories, Cramer's Phi coefficient was used, with cut-off points of 0.3 (medium) and 0.5 (large) effect as standards. The kappa measure of agreement was used to determine agreement between the two instruments (the UKIQ against the reference standard). A value of 0.5 for kappa represents moderate agreement, 0.7 good and 0.8 excellent.⁴²⁻⁴⁶ All were tested for statistically significant associations.

A patient global impression of change (GIC) question was included in the follow up questionnaire and this was used as the gold standard to assess clinically significant change over time,⁴⁷⁻⁴⁹ or responsiveness. The questionnaire was completed by mothers pre- and post-treatment and compared to the GIC. The correlation was calculated between the changed score for each question, as well as the corrected changed total score of the questionnaire overall.

Results

In all, 294 mother/infant dyads were recruited into the study with the baby's mean age of 8 weeks and the mother's mean age of 31. The infants were presented for crying (21%), feeding problems (20%), inability to sleep supine (19%), other sleeping problems (16%), head shape (8%) or check-up/difficult birth (16%).

Qualitative phase: The qualitative phase of the project gave domains which were infant behaviors (feeding, crying, sleeping, pain, movement patterns and abilities) and maternal feelings (anxiety, depression and quality of life). These were translated into questions that could be scored relative to the degree of the problem.

Phase 2: Development of the questionnaire: This resulted in a 12 question intake and follow-up questionnaire with the same questions. The follow up questionnaire also included a global impression of change (GIC) question used as the gold standard to assess clinically significant change.^{48,49}

Phase 3: Face and content validity: A total of eight participants were included, six experts in pediatric care and two experts in research, who agreed relevance, merit, content and face value of the instrument. Seventeen mothers found it relevant and useful.

The UK Infant Questionnaire was then presented to 20

Table 2. UKIQ Questions and Gold Standard Reference Measures

No.	UKIQ question	Gold standard/ surrogate measure	Why the gold standard/ surrogate measure was used
1.	Over the past few days, on average, have you considered your baby's feeding to be a problem?	Infant behavior diary	- Feeding easily correlated with 24 hour behaviour diary (Barr et al. 1988) - Eating Behavioural Questionnaire only validated for children (CEBQ), not for infants (BEBQ)
2.	Over the past few days, on average, have you considered your baby's sleeping to be a problem?	Brief Infant Sleeping Questionnaire (BISQ): "Do you consider your child's sleep as a problem?"	Research findings provide psychometric, clinical, and ecologic support for the use of the BISQ as a brief infant sleep measure for clinical and research purposes (Sadeh 2004)
3.	Over the past few days, on average, have you considered your baby's crying to be a problem?	Infant behavior diary	Infant behaviour diary is the gold standard and is easily correlated with the 24 hour behaviour diary (Barr et al. 1988)
4.	Over the past few days, on average, how much did your baby cry?	Infant behavior diary	- Infant behaviour diary is the gold standard for crying and an easy tool to measure crying quantities (Barr et al. 1988) - Only one other validated instrument: QUIC, which provides valid information about infant crying, but has not been widely disseminated and utilized (Miller and Green 2011)
5.	Over the past few days, on average, how easy or difficult has it been to console (comfort, calm) your baby when he/she cried?	FLACC (Face, Legs, Activity, Cry, Consolability) behavioral pain assessment scale	- Validated scale to assess consolability amongst four other pain measures - Validated as a parent-assigned pain score, which has been used as proxy measures of pain for children, such as those with cognitive impairment (CI), who cannot self-report (Voepel-Lewis et al. 2005)
6.	Over the past few days, on average, how comfortable (settled, relaxed) has your baby been while lying on his/her back?	None	There are no validated instruments
7.	Over the past few days, on average, how would you rate your baby's discomfort or pain?	FLACC (Face, Legs, Activity, Cry, Consolability) behavioral pain assessment scale	- Validated for children between 2 months and 7 years of age or individuals unable to communicate their pain. - Assesses for pain in children - Validated as a parent-assigned pain score, which has been used as proxy measures of pain for children, such as those with cognitive impairment (CI), who cannot self-report (Voepel-Lewis et al. 2005)
8.	Over the past few days, on average, how anxious (worried) or distressed (upset) have you been feeling about your baby's behavior?	Edinburgh Post Natal Depression Scale (EPDS): "I have been anxious or worried for no good reason"	The EPDS is the most widely used well-validated 10-item questionnaire designed to screen post-natal depression in mothers (Cox et al. 1987) - It has been validated in thirty-seven different research papers up to 2008 (Gibson et al. 2009)
9.	Over the past few days, on average, how depressed (feeling down, sad) have you been feeling?	Edinburgh Post Natal Depression Scale (EPDS): "I have felt sad or miserable"	The EPDS is the most widely used well-validated 10-item questionnaire designed to screen post-natal depression in mothers (Cox et al. 1987) - It has been validated in thirty-seven different research papers up to 2008 (Gibson et al. 2009)
10.	Overall, how would you rate your experience of motherhood and quality of life with this baby?	The short version of the World Health Organization Quality of Life questionnaire (WHOQOL-BREF): "How would you rate your quality of life?"	-Validated for postnatal mothers (Webster et al. 2010) - The WHOQOL-BREF is well-accepted and valid instrument for new mothers and can be utilized in postnatal clinical settings or for research purposes
11.	Over the past few days has your baby turned his/her head freely to both sides?	None	There are no validated instruments
12.	Over the past few days, on average, how much tummy time has your baby had?	Infant behavior diary	There are no validated instruments
13.	Since the beginning of treatment at this clinic, how would you describe the change (if any) in your baby's condition and or behavior?	Patient Global Impression of Change (GIC) scale	The GIC is the gold standard to measure clinically significant change in adults (Hurst and Bolton 2004)

mothers to complete. The average time to complete the questionnaire was five minutes. All participants were content with the length, wording, order, layout and content of the questionnaire and no changes were made at this stage.

Phase 4: Reliability: The results of the Intra-Class Correlation (ICC) coefficient indicated excellent test-retest reliability for each individual question. The overall ICC value of 0.96 (n=29) indicated excellent test-retest reliability of the entirety of the UK Infant Questionnaire.

Homogeneity (internal consistency): Cronbach's α was higher than 0.8, showing good internal consistency and that the instrument taps on different aspects of the same attribute.

Test-retest reliability: After test-retest and homogeneity testing, the UKIQ was considered reliable. The corrected item total correlations test showed that questions 1 — 10 can be added to form a total score, but items 11 and 12 did not contribute to the overall score. It was decided *a priori* that two questions would not be added into a total score. Question 11 (cervical spine rotation) was an untested question because of the absence of a gold standard comparison. However, it can be considered to have face validity based on maternal and expert opinion. Question 12 asks for specific times of prone play, rather than a rating scale, and therefore, is dissimilar in formatting to the first ten questions. Both are clinically important, but do not contribute to a total score.

Phase 5: Validity: Of the 12 questions tested, 10 were confirmed as valid to a statistically significant degree (Table 3). (Item 1 (feeding) correlated with the diary (Spearman's ρ .729). Question 2 (sleep) correlated with the Brief Infant

Sleep Questionnaire (BISQ), showing good agreement. Items 3 and 4 (crying) correlated with the diary (Spearman's ρ .568 -.612). Items 5 (crying), and 6 (pain) measured good agreement with the Face Legs, Activity, Cry, Consolability (FLACC) pain questionnaire. Question 9 (depression) indicated good agreement with the Edinburg Post-Natal Depression Scale (EPDS) at .737 (Cohen's Kappa) and Cramer's Phi (.780). Likewise, question 10 (maternal quality of life) correlated at a good level of agreement with the World Health Organization Quality of Life (WHOQOL) questionnaire (Spearman's ρ .667 and Cramer's Phi .514). Question 12 (prone play time) correlated with the diary (Spearman's ρ .688). Item 13 (UKIQ GIC) on the follow-up questionnaire showed high agreement with the existing validated Patient Global Impression of Change with Spearman's ρ .905.

Two items (question 7 — maternal anxiety and question 11 — cervical spine rotation) cannot be considered validated, as question 7 had only divergent validity (discrimination) with its reference standard, and question 11 has never been previously tested and therefore has no gold standard reference. Question 7 was removed from the UKIQ. Question 11 was considered important by both mothers and experts and therefore was retained, although it cannot be added to a total score. Item 12 cannot be summed either, as it is reverse scored from the rest of the questions (this was done to improve understanding by the mothers and to obtain clinically important information). The first ten items can be used as a sum of scores and each item can be used individually. The scores showed no apparent floor or ceiling effect.

Discussion

The 12-item UKIQ was developed through a systematic process of literature reviews, qualitative research, expert review, and pilot testing using 294 mother-infant dyads. Pa-

Table 3. Results of validity testing

Item	External measure used for correlation	Spearman's R	Cohen's Kappa	Phi (2x2)	Cramer's Phi
1	Infant behavioural diary	.729*			
2	Brief Infant Sleep Questionnaire (BISQ)	.641*	.476*		.470*
3	Infant behavioural diary	.612*			
4	Infant behavioural diary	.568*			
5	FLACC [^]			.678*	
6	FLACC [^]	.541*			
7	FLACC [^]	.688*			
8	EPDS& (anxiety)			.388(0.07)	
9	EPDS& (depression)		.737*		.780*
10	WHOQOL-BREF+	.667*	.410*		.514*
11	Cervical spine (validity not tested)				
12	Infant behavioural diary	.688*			
13	Patient Global Impression of Change	.905*			

* Correlation is significant at the 0.01 level or better; [^]Face, Legs, Activity, Cry and Consolability behavioural pain assessment scale; &Edinburgh Postnatal Depression Scale; +World Health Organization Quality of Life Questionnaire (abbreviated)

tient Reported Outcomes Measures (PROMs) for children are rare, and those that do exist are of low quality and lack reliability, validity or both or only measure a single variable.²⁵

Because of maternal concern for their infant's problems, along with the risk for persistence of those problems when inadequately addressed, the purpose of this project was to develop and investigate a questionnaire to follow progress during infant clinical practice. At first, the Bournemouth Questionnaire (BQ) was used as a model. However, this was abandoned to set the questions because of the need to use maternal interviews⁵⁰ (qualitative research) as a starting point, whereas the BQ had used a literature search as a basis to determine domains. However, their process for testing the questions was retained. Content validity was addressed by the implementation of current research⁵⁰ which advises including the parent and child into health care decisions. Thus qualitative research was used as the foundation for the instrument developed. The goal was to establish content validity that best served the target patient population by implementing their own needs and views. As such, the UKIQ is the only known pediatric questionnaire based predominantly on the views of the subjects it addresses, and this may be considered its strength.

This is balanced by significant weaknesses. Qualitative research has been criticized for lack of rigour.⁵¹ Further, it can lack generalizability. That is a distinct problem with this sample, as all subjects had presented to an outpatient chiropractic teaching clinic, and therefore were not necessarily representative of the general population of infants in the UK. However, the pediatric population in this clinic has previously been found to be representative of the broader infant population.⁵² Further, validating each question against its own gold standard reference is unique as was reliance on the infant behavioral diary. Although the behavior diary has been tested and is a gold standard, it has been noted by many researchers^{22,53} that the diary is onerous to complete and causes high drop-out rates. This is balanced by the significant rigorous testing of the infant diary and it remains the only gold standard in infant behavior today.²²⁻²⁴

Despite its drawbacks and need for further testing, relying on maternal views has led to a broad spectrum questionnaire which can be used clinically to understand the individual infant's health status and response to care in a well-child practice. It can provide a starting point, testing for clinical utility and pragmatic use in daily practice. Chiropractic care has been highly criticized for paucity of research in the infant patient.^{15,54} A reliable, valid and easy to use questionnaire as a replacement for the onerous gold standard behavior diary may go some way to encouraging more research due to the ease of measuring outcomes. The

aim was to allow both clinicians and researchers to integrate key aspects of the infant health parameters into their studies and practices. As such, the UKIQ is simply a first step to open the wider discussion between clinicians and researchers to continue development toward a widely useful instrument.

Meanwhile, this outcome measure may be useful to further the evidence base for infant care by chiropractors, which is urgently needed. It may also be useful for individual clinicians providing infant care to track progress with patients in their own clinic. The routine use of PROMS in clinical practice is widely advocated as a means of supporting patient-centered care, informing decisions and driving service quality.^{19,55}

A unique aspect of the UKIQ is that it can be used to assess the infant's progress relative to current public health issues of supine sleep, breastfeeding, and positional head deformation. As such, it tests the specific and broad concerns of the mothers for their child in well-patient care. Establishing efficacious treatments for infants facing difficulties in these areas is extremely important, not only for the health of the individual, but for their families and the broader community including the already economically stretched health services.

Conclusion

The UKIQ is a new parent-reported outcome measure for use with the infant patient in well-child practice. It was founded in maternal views through qualitative research, and demonstrated face validity, reliability, and validity as a clinically useful tool. As such, it provides a starting point for more investigation and discussion for further development. The UKIQ may be used in clinical practice by individual clinicians to monitor progress of infant patients, and could also be used in future outcomes focused research for this age group, as well as community surveys to sample maternal concerns. The practical and clinical utility with widely varied populations along with clinical significance of scores requires further study.

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Evaluation and treatment of breastfeeding difficulties associated with cervicocranial dysfunction: a chiropractic perspective

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ABSTRACT

Increasingly, parents are presenting to the chiropractic office with the chief complaint of breastfeeding dysfunction. Early and consistent breastfeeding support is paramount to the dyad's ability to establish a functional breastfeeding relationship. It is critical that chiropractors treating this population recognize feeding dysfunction and understand the mechanics of breastfeeding, what might alter those mechanics, and the importance of the role of the IBCLC in supporting the restoration of functional breastfeeding after the chiropractic adjustments have reduced the NMSK dysfunction. This paper outlines some of the mechanical dysfunctions that might interfere with the normal transfer of milk for a neonate.

Keywords: Chiropractic, breastfeeding, breastfeeding difficulties, biomechanics, pediatric, neonate

Introduction

Increasingly parents are presenting to the chiropractic office with the chief complaint of breastfeeding dysfunction. Through the efforts of organizations like La Leche League International^{1,2} and the International Association of Tongue Tie Professionals (IATP),³ books^{4,5} and professional publications,⁶⁻¹⁵ and a boom in activity on social media of both professionals and concerned parents,^{16,17} The International Board Certified Lactation Consultants (IBCLC) and other healthcare providers (such as midwives, nurses in the hospital, pediatric office and homecare settings, and pediatricians) who support the nursing dyad are becoming more familiar with and adept at recognizing neuromusculoskeletal (NMSK) dysfunction that might interfere with breastfeeding.

As has been previously described,¹⁸ it is important that early and consistent breastfeeding support be implemented to assist the dyad's ability to establish a functional breastfeeding relationship. Therefore, chiropractors working with this population should understand the mechanics of breastfeeding, what might alter those mechanics, and recognize the importance of the IBCLC in supporting the restoration of functional breastfeeding after the chiropractic adjustments have reduced the NMSK dysfunction.^{6,10} The purpose of this paper is to outline some of the mechanical dysfunctions that might interfere with the normal transfer of milk for a neonate.

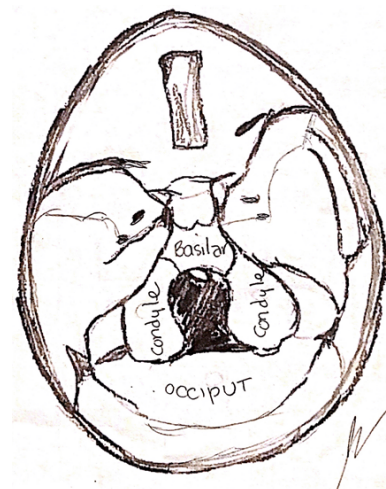
The Craniocervical Junction

The anatomy of the craniocervical junction (C01) is an important consideration in the neonate who is not feeding functionally at the breast or on the bottle. The skull devel-

ops from viscerocranium and neurocranium, which develop into the facial bones and the portion of the skull that protects the brain. The neurocranium consists of the chondrocranium and dermatocranium which give rise to the skull base from endochondral ossification and the calvarial vault from membranous ossification respectively. The rapid growth of the neurocranium is stimulated by expansion of the size of the brain over the first 7 years of life with fontanelles closing (at different times) over the first two years of life.

The occiput, not yet fused at birth, is in 4 segments^{19,20} which can be displaced (or misaligned) by the normal compression of the cranium during the birth process. They, like other plates of the cranium, may misalign or overlap and remain so, when subjected to inordinate or prolonged forces.

Figure 1. The neonatal occiput



The lateral components of the occiput each possess a condyle that articulates with the first cervical vertebra (atlas or C1) at the superior articular surface of the lateral mass. The normal movements between the occiput and the atlas (C01) are: flexion and extension (nodding the chin to chest and back at the cranial base allowing an infant to lift their head when lying supine or tip their head back to adore their parent when resting on their shoulder, or the motion required to shake the head “yes”), rotation (shaking the head “no”) and lateral flexion (ear to shoulder) along with the possibility of an anterior, posterior or lateral translation of the full cranium on the atlas. Although subtle, identification of restricted movement in one of these planes helps determine the site of subluxation. Restricted movement can be due to inflammatory changes, misalignment of the articular structures, ligamentous injury, muscle spasm, tension in the fascial planes or asymmetry in development. It can also occur as a result of compensatory mechanisms put in place by the infant due to other physiologic, neurologic or structural obstructions to the normal mechanics of feeding.²¹

Any misalignment of the condyles can interfere with function in a direct and obstructive way, functionally acting like a rubber wedge carefully placed under a door to prevent it from closing. Indirectly, if the misalignment results in nociceptive input (caused by a myriad of alterations ranging from fascial tension to an alteration in the patency of one of the foramen through which cranial nerves pass), it could also lead to a decrease or increase in muscle tone, the latter perceived as muscle tension or spasm. Change in muscle length or activity can adversely influence a functional joint. But when this involves muscles that have been demonstrated to bridge to the dura, via the fascia (like the rectus capitis posterior minor or the rectus capitis posterior major), nociception may be an even more critical factor in the ongoing dysfunction. Torsion or strain at the myodural bridge has been associated with cervicogenic headaches in adults and could potentially cause the same type of neurogenic pain for a neonate, as well as perpetuating the pain spasm cycle.^{22,23} If the condylar displacement affects foraminal patency for the cranial nerves, other systemic symptoms may ensue (for example, compression or traction of the fascial sleeve of the vagus nerve (CN X), can result in an alteration in vagal “tone” or autonomic dysregulation which could affect heart rate and blood pressure, respiratory rate, colic-like symptoms, swallowing — and the suck-swallow-breath synchrony — and cause an alteration in gut motility and excretion).²⁴

If movement is altered at the level of the occiput and atlas, the brain will quickly initiate compensatory action of accessory muscles to attempt to accomplish important functions like breathing and eating. Breathing under stress or physical exertion will be quickly assisted by engaging the sec-

ondary muscles of respiration in the cervical spine. Feeding (particularly to secure the mouth around the nipple) will be supported by these same muscles as well as the submandibular muscles, the muscles of mastication and the muscles of the lips (primarily the orbicularis oris). The recruitment of these secondary muscles can result in functional “postural” changes (rounding of the shoulders, posterior translation of the cranium on the atlas, flexing the head on the chest, tilting of the head if unilaterally recruited, etc.).

Next let us consider how two examples how alteration in function of musculature can influence the mobility at C01.

The styloglossus, the shortest and smallest of the three styloid muscles, arises from the anterior and lateral surfaces of the styloid process near its apex, and from the stylomandibular ligament. The styloglossus is innervated by the hypoglossal nerve (CN XII), it functions to draw up the sides of the tongue to create a trough for swallowing and retract the tongue.¹⁹

The stylohyoid muscle arises from the posterolateral surface of the styloid process of the temporal bone, near the base. It is a slender muscle, lying anterior and superior of the posterior belly of the digastric muscle and inserts onto the body of the hyoid bone at its junction with the greater cornu. A branch from the extracranial path of the facial nerve (CN VII) innervates the stylohyoid and the muscle functions to retract the hyoid and elevate the tongue.¹⁹

Figure 2: The stylohyoid and styloglossus muscles



If C01 is subluxated (fixed or immobile in any or all planes of motion), when either of these muscles contracts, the action of the muscle will be altered by the restricted mobility of its proximal attachment (the styloid process). This will result in increased movement of the distal attachment as

demonstrated by retraction of the tongue or elevation and retraction of the hyoid bone. Likewise, if the tongue is tethered to the floor of the mouth or the movement of the hyoid is restricted by muscle activity or fascial tension, the proximal attachment (the styloid process) will be pulled towards the distal fixed attachment and affect the alignment of the cranium on C1 (permitted by the mobility allowed at C01 at the condyles).

Visible postural alterations and associated compensations due to dysfunction of the craniocervical junction can range from the subtle: slight head tilt, preferential rotation, chin to chest posture or the presence of a wry smile (as the mandible deviates) to more obvious distortion like torticollis or plagiocephaly as a result of repetitive or sustained asymmetric contraction over time. If unaddressed in infancy, postural changes can manifest in toddlerhood as toe walking, wide-based stance or gait, and increased AP curves of the spine as the continued compensatory recruitment disrupts the development of proprioceptive systems that determine position and balance for the toddler.

It is this author's hypothesis that biomechanical restriction beginning with constraint or compensatory development of asymmetry in the womb could potentially be significant enough to negatively impact the normal egress of the fetus during labor resulting in potential compromised situations like an asynclitic presentation or shoulder dystocia because of the lack of full range of motion during the spiral descent. Other potentially far-reaching physiologic effects of altered biomechanics in infants and children can include failure to attain developmental milestones (due to decreased range of motion, for example), irritability, mouth-breathing, snoring, sleep apnea and disordered sleep,²⁵⁻²⁷ feeding challenges at breast or by bottle^{4,6} and potential alterations in cranial nerve function which could result in dysregulation of respiratory, cardiac and or digestive function.²⁸

Assessment of the cranial nerves is critical, as altered cranial nerve function may be the only indicator of articular dysfunction at C01. For example, vagal irritation (due to its proximity to the occipital condyle as it exits the skull through the jugular foramen) could potentially result in gastrointestinal distress or affect the function of cranial nerves V, VII, IX, X and XII and result in failure to root, suckle and swallow.

Manual Technique to Correct Dysfunctional Vertebral Segments

Chiropractic evaluation specifically identifies the areas of dysfunction and the chiropractic adjustment addresses it by restoring articular integrity, whether at the craniocervical junction or associated articulations. Both evaluative and adjusting techniques should be modified based on devel-

opmental anatomy, dictated by the neonate's gestational age and concomitant conditions. Todd et al, published a review of the literature describing the level of force applied in a chiropractic adjustment or spinal manipulative therapy (SMT) for a pediatric patient based on age. A set of guidelines for different age groups has been developed by Marchand, "drawing on findings from an extensive study of tensile strength and osteoligamentous failure rates in pediatric spines as well as a report of transient bradycardia and apnea events that occurred with thrusts of 50 N to 70 N in infants younger than 3 months. Marchand has recommended that SMT be applied at a maximum cervical loading of 20 N for neonates."²⁹⁻³¹

A chiropractic evaluation should be multidimensional and should include a clear image in the chiropractor's mind of the anatomy of the pediatric patient along with his or her active and passive assessment of range of motion and visual assessment of posture and attitude coupled with listening for altered or strained breath sounds, clicking of the tongue, swallowing or sucking in of air, the sound (and sometimes smell) of the burp, hiccup or flatulence (or stooling) are just a few of the sensory impressions that a chiropractor will integrate with the infant's history to differentially diagnosis and plan a therapeutic approach.

Evaluation of the infant should begin with a soft tissue evaluation appreciating the connective tissue components that can interfere with joint mobility and function. Manual techniques employed are too varied to cover in this paper. Many current chiropractic techniques include soft tissue release to facilitate mobilization. Extensive study of fascia "based on functional relevance of myofascial chains" is recommended by Wilke, et al. (2016) in his systematic review.³² An appreciation of the anterior and posterior superficial lines and anterior deep lines as outlined by Myers³³ and applied to the infant with breastfeeding dysfunction might prove to facilitate shorter, more effective courses of treatment.

Manual assessment of each of the individual cranial bones would be a part of the comprehensive assessment of the infant with breastfeeding difficulty. For the purposes of this paper, we will limit our discussion to the occipital bone, but a comprehensive assessment is holistic and will assist the practitioner in evaluating the intimate balance of mobile articulations and the soft tissue that both moves and supports them.

Palpation of the occiput will include the evaluation of all the aforementioned ranges of motion and the motion of the occiput as it articulates with the sphenoid at the sphenobasilar junction. This junction normally flexes and extends with respiration, but may be fixed in flexion, extension, lateral flexion or torsion. Palpation should also include evalu-

ation of the individual segments of the occiput (anterior, posterior and lateral condylar segments) to assess medial and lateral translation or internal and external rotation, as well as anterior and posterior translations of the individual segments. (Figure 3).

As stated earlier, all chiropractic techniques taught in the academic setting are modifiable for gestational age and anatomy. For example, methods of choice for this author are cranial adjusting (for example, as taught by DeJarnette^{34,35} and Upledger³⁶), low velocity, low amplitude (low force) (manually (digitally) or with a pediatric drop head piece), percussive instrumentation (like the Activator Technique)³⁷ delivering a measurable, short range, reproducible force, or non-force techniques maintaining steady digital contact (“press and hold”) as a fulcrum about which a neonate moves until they release the restriction at their own pace. In the press and hold technique, the emphasis is on acting as a stationary fulcrum and letting the neonate “lead” the myofascial release which ultimately restores vertebral motion. This avoids force or excessive range-induced trauma and reduces the risk of tissue injury and activation of the sympathetic response which can lead to autonomic dysregulation if the trauma exceeds the neonate’s adaptive threshold. The author refers the reader to the Marchand article that reviews quite a number of chiropractic techniques and age-appropriate modifications beyond the scope of this commentary.²⁹

Gently supporting the head with fingertip contact along the occiput using two fingers to assess the “spread” and position of the condyle or using all fingers to encourage anterior translation or extension at C01 will gently accomplish mobilization, often without distressing the neonate. At times, they will express intermittent distress as they “unwind” the fascia (connective tissue components that may have been injured during the birth process or from misuse) or if a trigger point in a suboccipital muscle is compressed. Patience and a consistently firm but gentle digital contact usually al-

lows them to work through the discomfort until they are relaxed and at ease, often falling asleep as the parasympathetic system becomes more dominant than the sympathetic system that had been engaged when all systems were not functioning properly.

Developing a discerning touch involves understanding the neonatal anatomy. For example, that the occiput has four individual pieces with each segment’s position and mobility influenced by its own fascial envelope (or as mentioned earlier, from a distal restriction along one of the associated myofascial trains). The misalignment of one segment may subtly change or be completely released by effectively releasing the soft tissue or another subluxated segment. Consider releasing the taut fascial envelope and use good clinical judgment and develop precise manual skills before applying any direct force (no matter how modified) to a motion segment. In most cases, inducing nociceptive input is more likely to cause more restriction than less.

Highly developed observation and palpation skills; familiarity with anatomy and function; discernment; clinical experience; appreciation of the role of myofascial components in dysfunction; and a skilled, specific adjustment are the tools that will allow the chiropractor to facilitate normal motion and neurologic regulation for the neonate demonstrating breastfeeding difficulties, interference with the suck-swallow-breath synchrony or potentially other physiologic challenges.

Conclusion

Although only one articulation was specifically, and not necessarily exhaustively discussed in this paper, the implication that multiple articulations and myofascial components may be compromised is implicit in this discussion. Therefore, it is important that the neonate experiencing any feeding (or breathing) challenges should also be evaluated for NMSK dysfunction and be sent for evaluation by a qualified IBCLC.

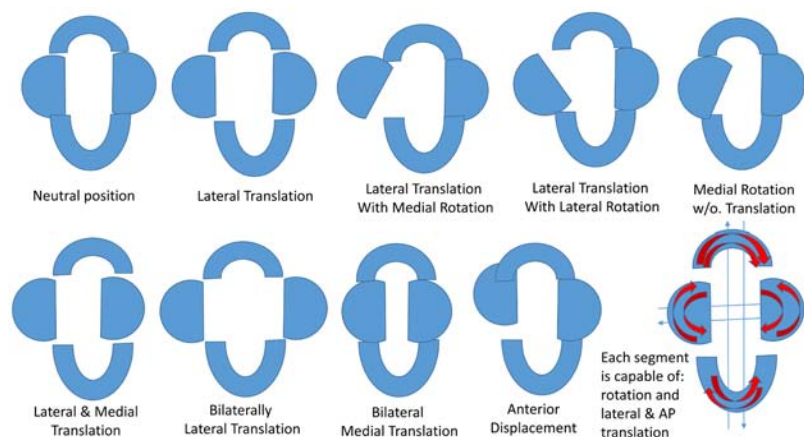


Figure 3.

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What is Tummy Time: is it necessary for newborns?

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Introduction

Chiropractic offices are receiving large numbers of infant patients into their practices. Consequently, new guidelines have been forthcoming to assist chiropractors in applying their best evidence practice to this age group.^{1,2} However, very little has been said in how we give advice, particularly in the important topic of prone play (“tummy time”) for the infant. It is not always recognized that research data must be translated into clinical practice in order for the practical applications to benefit the end user, the patient, and in this case the parent and the infant. Why would such a simple concept, such as an infant playing in prone position be an issue for discussion between the doctor and parents? It is because the simple messages have become convoluted, confusing and difficult to follow. It is perhaps time to go back to the beginning.

What is the postural problem?

Most new mothers report little, if any physician advice on daytime positioning, but 90% report receiving information on infant sleep positioning.³ In general, correct positioning of the neonate is very important for postural control for any infant at risk of developing fixed deformities, but little specific advice may be given by physicians, except for the “Back-To-Sleep” program.⁴

The long-term 1992 advice from the American Academy of Pediatrics recommending that caregivers always place a baby on his or her back for sleep at night has come through loud and clear.⁴

This advice has been frequently reiterated, “reaffirmed” (what the AAP does periodically to keep specific policies in the public eye) and often repeated that the supine positioning has had a positive reduction in SIDS (sudden infant death syndrome).^{5,6-10} This advice may or may not stand up to scrutiny and is a potential and necessary topic for extensive evidence based review.¹¹ That review cannot be done in the context of this paper. Our role and responsibility is to provide resources for parents so that they may attempt to make informed decisions based on the available evidence.

Another effect from Back-To-Sleep, and the topic of this commentary, has been a common occurrence of misshapen heads in the pediatric population (Table 1).⁵

Why does head shape change with supine sleep? Since

Table 1. Prevalence of head shape deformity in infants

Age	Percentage
6 weeks	16%
4 months	19.7%
8 months	9.2%
12 months	6.8%
24 months	3.3%

most of the infant’s time is spent in sleep, if the head position doesn’t vary, the soft skull is likely to flatten from consistent, weight bearing positioning. The problem is that the more it flattens, the harder it is for the infant to change position. The more time passes in only one position, the flatter the malleable, weight bearing area becomes, losing the roundness of the cranium and resulting in head deformation, or what is termed “plagiocephaly” or misshapen skull. This can occur on the posterior aspect of the skull, which is termed “brachiocephaly”, or very commonly, unilaterally (only on one side) when an infant has preferred or restricted neck rotation.

Although head shape problems do decline with age, there are still a significant number that suffer from longer term problems. Research suggests that infants with positional plagiocephaly are at risk of a delay in motor milestones.¹²⁻¹⁷ Although many physicians term the cranial deformation “purely cosmetic,” it may be that cosmetic head deformity is merely a marker for the problem, not the real problem, which is more likely the lack of opportunity to play in prone position which supports development.¹⁷

Chiropractors will be very familiar with the problem of the infant who cannot rotate his or her cervical spine equally to both sides (due to vertebral fixation or muscular imbalance), thus causing a flatness on one side of the cranium (more commonly the right side).¹⁸ Once the infant has settled into this preferred posture, the more the head flattens, the more difficult it is to overcome gravity and the more staying to that side becomes a habit, profoundly preferred by the child. Parents may accept this as typical behaviour and the head shape may not even be noticed by them and it is often first pointed out by the doctor on the 8 week wellness exam. If the baby is not prescribed and administered treatment to

allow him/her full range of motion of the cervical spine and is not given good instruction on the need for tummy time, the problem may persist. There is some evidence that chiropractic care is helpful to reduce the head deformity through improving the infant's ability to rotate equally in both directions.¹⁸ The AAP recognized that infants were missing out on important health motor develop by 1996 and recommended to parents that they vary their infant's head position in their sleep and give them some tummy time when awake, beginning at birth.⁶ Unfortunately, this advice was largely ignored.³

Despite that early warning about the need for tummy time, it has never really become common advice for health care professionals to disseminate. Parents have had to learn on their own of its many benefits and they sometimes have a fear of placing their baby on his/her front based on the 1992 information linking any position except supine with SIDS.⁴ Many parents did not realize that supine is the position for sleep, but that day-time position should be varied for the health of the child.

What is it important for parents to know and understand?

Environment makes a difference to baby's development and babies need to move within their environment. The authors are proposing a guideline for parents to use in order to help them understand how much tummy time may help their babies adapt to their environment in a positive way and to prevent or help alleviate any positional head defor-

Age	Minutes/day
1 week	5 minutes/day
4 weeks	10
8 weeks	20
12 weeks	45
16 weeks	80 minutes/day

mation caused by continuous supine positioning (Table 2). Remember that the time per day can be split into smaller time intervals throughout the day. Make it fun with toys and interaction, so that they gradually get used to it and learn to enjoy the time and the new point of view. It is key to remember that until or unless the AAP guidelines are reviewed and refuted with evidence based information, lay babies down to sleep on their back, do not lie babies prone on less than firm surfaces (not on pillows, comforters, pillow tops, etc) and always supervise the tummy time. Doing these things may lower the risk for both SIDS and developmental problems.

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Positive chiropractic treatment outcome of migraine without aura in a 6-year-old presenting with sleep bruxism and chronic sinus congestion: a case-report

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ABSTRACT

Migraine is a common disorder affecting up to 10% of children which can interfere with school attendance, academic and sport performances.^{1,2} The proposed pathophysiology has evolved from one purely vascular in origin to perhaps a neuro-inflammatory etiology.^{3,4} Clinical evaluation is the mainstay of diagnosis and should include family history, sleep habits and environmental triggers.^{5,6,7} Spinal manipulation of the cervical spine in adults and teenagers has shown similar effectiveness as prophylactic medications.^{8,9} A Canada-wide survey revealed that 7% of chiropractic pediatric patients presented for headaches.¹⁰ Unfortunately, there is no substantial data at this point to support the benefit of spinal manipulation in the pediatric sufferers of migraine.^{11,12,13} Recent research found a link between sleep parasomnia (bruxism), colic and migraine sufferers.^{7,14,15,16} To achieve a precise diagnosis, clinicians must refer to the latest diagnostic criteria to differentiate true migraines from tension headaches, sinus headaches or other organic causes.¹⁷ This case report discusses the chiropractic treatment outcome of a 6-year-old female patient suffering from migraine without aura, sleep bruxism and chronic sinus congestion. The patient was treated with mechanical assisted and manual manipulation techniques, massage and sinus lymphatic drainage. After 4 visits migraine intensity reduced from a 5/10 to a 2/10 and the frequency of headaches were reduced from 3 times per week to once a week without the use of medication. After 6 weeks of care, patient symptoms were significantly reduced but the treatment plan was not completed because the family relocated and continued travel to the office was prohibitive. After 17 weeks, the patient relapsed and 3 weeks later, they resumed care with progressive improvement of the child's well-being once we eliminated migraine triggers such as sun exposure, incorporation of resting periods with naps in the afternoon and cardiac coherence training exercises before bedtime.^{1,5,7,18,19} This case report highlights the need for high quality research on the effect of spinal manipulation in the treatment of pediatric migraine to provide a potential alternative option to the traditional medical treatment of migraine. This case also opens the discussion on the benefits cervical manipulations and cranial work may potentially have on sinus drainage and immune system response in autonomic conditions like sinus congestion.

Keywords: Migraine, children, spinal manipulation, bruxism, sinusitis

Introduction

Following tension headache, migraine is the second most common cause of chronic recurrent headache in school children with a prevalence ranging from 3.2 to 14.5%.¹⁴ Pediatric migraine differs from adult migraine in that they are typically of shorter duration and bilateral in children.³ Di-

agnostic criteria are based on the international classification of headache disorders 3rd edition (ICHD-3 beta) from the international headache society (Table 1).²⁰

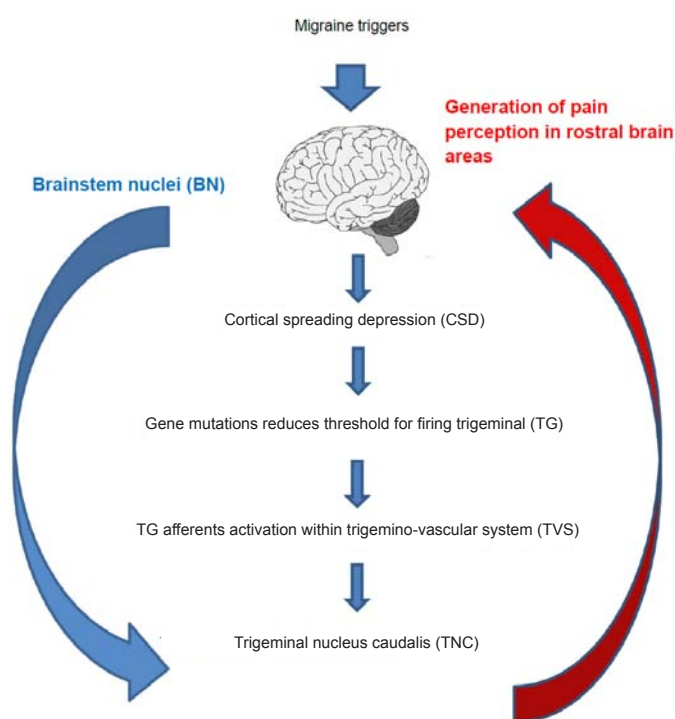
Migraine is divided into six major categories, the two most important of which are migraine without aura and mi-

Table 1: International headache society diagnostic criteria for pediatric migraine without aura^{3,14,20}

- A) A minimum of 5 attacks fulfilling features B to D
- B) Headache attack lasting from 2 to 72 hours
- C) Headache has at least 2 of the following 4 features:
 - (1) Unilateral (frontal/temporal) location commonly bilateral in young children
 - (2) Pulsating quality
 - (3) Moderate to severe pain intensity
 - (4) Aggravated by or causing avoidance of routine physical activity
- D) During the headache at least one of the following:
 - (1) Nausea and/or vomiting
 - (2) Photophobia and phonophobia (may be inferred from their behavior)
- E) Not attributed to another disorder

graine with aura.³ According to the literature, children with migraine have a genetic predisposition in some way activated by an environmental or physiological stimulus such as exposure to drugs, diet, stress or puberty.^{3,6} The pathophysiology of migraine in children is not perfectly understood due to a dearth of research in the field.¹¹ Recent studies using PET, intracarotid SPECT (single-photon emission computed tomography), MRA (magnetic resonance angiography), functional MRI and TMS (transcranial magnetic stimulation) concluded that the pathogenesis of migraine attacks is explained by a combination of vascular and neurogenic events. It has been proposed that the activation of central cortical and brainstem pathways in parallel with the peripheral trigeminovascular system results in a release of neuropeptides such as CGRP (calcitonin gene-related peptide) and substance P inducing vasodilation and neurogenic inflammation.^{14,21,22,23} (Figure 1).

Figure 1: Migraine pain generation* ^{14,21,22,23}



*Migraine triggers initiate the neuronal excitation that leads to clinical manifestation in children with a genetic vulnerability to migraine. CSD triggers plasma protein extravasation from cerebral blood vessels, which in turn activates TG afferents within TVS. Gene mutations could reduce the threshold for firing TG neurons. Signals are transduced to the TNC, which receives modulatory inputs from other BN. The TNC projects to rostral brain areas, where the perception of pain is generated.

Cranial autonomic symptoms such as nasal congestion are frequently found in pediatric migraine, often leading to a misdiagnosis of sinus headache, subsequently delaying appropriate migraine treatment.^{17,24} Recent research

found links between sleep parasomnia (bruxism), colic and pediatric migraine sufferers.^{7,14,15,16} Few older randomized control trials (RCTs) and a more recent one presented by Tuchin suggests that chiropractic spinal manipulative therapy might be equally efficient as propranolol (beta blocker) and topiramate (anticonvulsant) in the prophylactic management of migraine.^{9,25,26,27} This case-report will discuss the chiropractic care of a 6-year-old female patient suffering from migraine without aura presenting with sleep bruxism and chronic sinus congestion.

METHOD

An online literature search was conducted using scientific journal databases; PubMed, Elsevier, Index to Chiropractic Literature and Google Scholar. Databases were searched from inception through October 2015 using the keywords: Migraine, children, spinal manipulation, bruxism and sinusitis. Publications utilized included systematic reviews, randomized controlled trials and case studies. Other articles and resources were used to provide background information.

CASE PRESENTATION

A 6-year-old Caucasian female patient presented with frequent migraines without aura for the past 6 months. The migraines were located in the frontal region bilaterally and occurred 3 times per week graded at a 5/10 on a visual analog pain scale. The patient described the migraine as a diffuse pounding sensation behind her forehead and she felt slightly nauseated. During the episodes the patient could usually continue most of her daily activities at the kindergarten with the exception of some physical activity that involved jumping. On occasion she had to miss school and did not attend her gym class. Parents have reported that the child often ground her teeth at night and although she dealt with chronic sinus congestion year round, it was more intense during the winter time. The patient's pediatrician recommended the use of ibuprofen when needed which alleviated the patient's symptoms. The patient was examined by her optometrist 4 months ago who ruled out any visual or ocular problem.

Medical history included mild colic around 2 to 3 months of age followed by chronic otitis media between 3 to 4 years of age. She received 5 rounds of antibiotics for the recurrent otitis media. At age 4, she developed recurrent sinus congestion, most intense during the winter season. Since the age of 5 she often suffered from constipation. The migraines started when she was 5 and a half and progressively became worse. Based on her intake form which includes a detailed description of typical breakfast, lunch and dinner, she has a balanced diet with home cook meals. Her parents denied attempting any food elimination diets and had not identified any specific food triggers. Sun exposure and fa-

tigue were described as common migraine triggers for their daughter. The patient rarely drank water and preferred juices. Both parents suffered from migraines at a younger age. Their home was a smoke-free and pet-free environment. Previous history of injury included a fall from her crib at 6 months of age, a recent fall while tobogganing, and a fall on her head later during the course of care. The patient was an active young female who enjoyed participating in ballet and soccer. Severe migraine episodes interfered with her participation in day camp and athletics. She would watch 1-2 hours of television per day and played on the Leap Pad 2-4 hours per week.

CLINICAL FINDINGS

Complete blood count (CBC), Glucose AC, TSH and Epstein-Barr antibodies (VCA IgG IF / EBNA IF) in addition to an eye exam were done 3 months after the beginning of the symptoms. Blood work and eye examination were unremarkable. Parents had been using a “Netty Pot” and administering “Avamys” nasal spray to clear sinus congestion when it was necessary. Ibuprofen was used 1-2 times per week on an as needed basis. On examination cervical passive and active range of motion (ROM) were within normal limits. Muscle tension was noted in the left sub-occipitals, temporalis, masseter and bilateral trapezius muscles. Lymph nodes palpation was sensitive in the posterior cervical chain bilaterally at the initial exam. Segmental dysfunctions palpated at C0-1, T1-2 and L1-2. The temporomandibular joint (TMJ) was palpated superior on the right side with ipsilateral joint restriction. Cranial bones analysis presented an inferior left sphenoid with increased muscle

tension at the frontal-temporal junction bilaterally. Postural analysis revealed a mild forward head carriage and bilateral excessive foot pronation. Gait, coordination, blood pressure and neurological testing was within normal limits.

DIAGNOSTIC AND ASSESMENT

The patient’s clinical presentation met the International Headache Society diagnostic criteria for migraine without aura (ICHD-III beta) (Table 2). The pulsating quality of the headache accompanied with nausea and the avoidance of physical activity led to a probable diagnosis of migraine rather than tension headache. According to Heyer and Pappetti, the PedMIDAS is the only instrument validated to assess migraine disability among school-aged children.^{28,29} This diagnostic tool presents questions to rate the impact of migraine on school performance and level of disability at home. The PedMIDAS score was rated at 4 (little to no disability) at the initial exam and 22 (mild disability) 17 weeks later while she was attending day camp 2 weeks after her 5th visit (Table 3).³⁰

Table 3: PedMIDAS grading scale³⁰

PedMIDAS Score Range	Disability Grade
0 to 10	Little to none
11 to 30	Mild
31 to 50	Moderate
Greater than 50	Severe

The analog pain scale was rated at a 5/10 at the initial exam to 8/10 as her symptoms relapse 17 weeks after the

Table 2 : Timeline

2009	Infantile colic
2012	Onset of chronic otitis media
2012-13	5 series of antibiotics for otitis media
2013	Onset of chronic sinus congestion
2013-15	Use of “Netty pot” and “Avamys” nasal spray to clear sinus Congestion
2014	Onset of migraines and recurrent constipation Complete eye exam, CBC, Glucose AC, TSH and Epstein-Barr antibodies (VCA IgG IF / EBNA IF)
03/2015	1st chiropractic consultation followed by 6 chiropractic adjustments in 2 months period
05/2015	Family moved to a new city an hour drive away
06/2015	Patient had 1 mild migraine per week for the past 2 months and less sinus congestion. She didn’t need to rely on medication. Preventive chiropractic care implemented at 1 visit per month.
07/2015	Return of migraines while attending day camp in their new city. Emergency check-up for intense migraines with abdominal pain present for the past 3 weeks. Elavil, naproxen and lax-a-day was prescribed with no improvement of the migraine symptoms.
08/2015	Chiropractic care was re-introduced at 1 visit per week for 1 month. Migraine episodes have reduced in intensity but are still present on a daily basis.
09/2015	Patient is on Naproxen and Elavil daily following doctor’s recommendation. Constipation is occasionally alleviated with lax-a-day. Patient receives chiropractic care every 2 weeks. Sinus congestion is well controlled with the use of the “Netty Pot” and parents have stop using the “Avamys” spray. Patient still grinds her teeth at night.
10/2015	Cardiac coherence training exercise was prescribed before bed time to promote relaxation. Patient rarely grinds her teeth at night. Patient doesn’t wake-up with migraine, mild headache starts later in the morning but does not interfere with school or sports performance. Patient doesn’t have any sinus congestion. Chiropractic care is maintained at 1 visit per 2 weeks.
11/2015	Patient was assessed by a neurologist who stopped Naproxen and Elavil. Medication should only be used during an acute episode. Patient has occasional mild headache, she performs well in school and athletics. Preventive chiropractic care implemented at 1 visit per month.

initiation of chiropractic care. The frontal location of the migraine was less suggestive of organic pathology, which is most commonly located in the occipital region.¹⁴ There were no red flags present suggestive of tumor, meningitis or infection. The prognosis for this patient was good given the family's strong genetic predisposition to migraine, mild disability level, early diagnosis of the condition and healthy lifestyle of the patient.

THERAPEUTIC INTERVENTION

The patient started chiropractic care 6 months after the initial onset of migraine symptoms. Treatment consisted of spinal manipulation at C0-1, T1-2 and L1-2 using Thompson's drop, Activator instrument and modified Diversified upper cervical adjusting techniques in addition to massage of the left sub-occipitals, temporalis, masseter and bilateral trapezius muscles. Gentle mobilization of the right TMJ, sinus lymphatic drainage and cranial technique to the sphenoid were also applied. The patient received 5 chiropractic treatments over 3 months. Chiropractic care was re-introduced 6 weeks after following the flare-up of migraine symptoms to a frequency of 1 visit per week for a month and 1 visit every 2 weeks for 2 months. No adverse reaction to chiropractic care was reported by the patient or parents during the course of care. In addition, the patient was recommended to perform daily neck stretching exercises, increase water intake to 4-6 glasses per day and reduce time watching television and playing with her Leap Pad. Parents were also advised to have their daughter go through skin prick testing as well as blood work IgG ELISA testing to identify any possible environmental or food allergies / sensitivity.³¹ At most recent follow-up this testing had not been performed. After 27 weeks of care (10th visit) the patient was instructed to do cardiac coherence training exercises (deep breathing) before bedtime to promote relaxation and restful sleep.^{1,19}

FOLLOW-UP AND OUTCOMES

During the first month of care the patient didn't take ibuprofen for migraine. The intensity of migraine dropped to a 2/10 on a visual analog pain scale and frequency reduced to 1 episode a week and no longer interfered with the patient's daily activity. The patient then had a relapse of her symptom 3 months after the beginning of chiropractic care which brought her to the hospital's emergency with a migraine rated at an 8/10 on a visual analog scale and accompanied with nausea and constipation. She was kept under observations for few hours and the attending prescribed Elavil, Naproxen and Lax-A-Day. The patient was dismissed from emergency and referred to a neurologist in the following 4 months. Three weeks after the relapse of her symptoms the patient's complaint was unrelieved by the medication and she resumed chiropractic care. The same treatment protocol was administrated to the patient at a frequency of 1 visit per week for a month then 1 visit every 2 weeks for

2 months. After 2 weeks of re-introducing chiropractic care the patient was still having migraines on a daily basis but they were reduced in intensity to a 4/10 and were alleviated with rest, reduced physical activity and sun exposure. Upon returning to school she reported only mild headache without pounding nature and she was able to fully participate in school and athletics. The parents also indicated that it was the first summer where she wasn't complaining of mild congestion after going to the pool or playing outside. The parents reported a significant reduction of bruxism at night and the patient reported feeling well rested when she woke up in the morning. The parents continued administering the "Netty Pot" preventatively. Consultation with the neurologist, shortly after the patient's 11th visit, revealed that the patient no longer needed Elavil and Naproxen on a daily basis and should only use the medication during acute episodes.

DISCUSSION

This case highlights other health conditions related to migraines and stresses the importance of a thorough intake and clinical examination. In order to achieve a precise diagnosis the clinician must refer to the International Headache Society diagnostic criteria for migraine ICHD-III beta and the PedMIDAS questionnaire.^{20,28,29} This latest diagnostic tool (PedMIDAS), is questionable in its accuracy when completed during the summer holidays since the first 3 questions are related to school performance.²⁸ Since the brainstem regulates vascular tone and nociceptive function in the body it is logical that upper cervical spinal manipulation could have a positive impact on migraines^{14,13,32,33} however there is little literature to demonstrate the effectiveness of spinal manipulation on pediatric migraine. Three out-dated RCT's on spinal manipulation and migraine were found in the literature review.^{9,26,27} A more recent RCT from Tuchin, Pollard and Bonello discussed the effectiveness of spinal manipulation in migraine patients ranging from 10 to 70 years of age.²⁵ 50% of the subjects of this study noticed a reduction in the intensity of the migraine episode. 80% of the participants reported stress as a major contributor of migraine. The research conclusion was that spinal manipulation can have a positive effect on stress related conditions like migraine. Only one case report from 2013 was found by Kuhn where functional neurology and spinal manipulation was applied in the management of a 15 year old boy with migraine and behavioral learning difficulties.³³ After 4 months of care the patient was no longer suffering from migraine, his vision improved and tics reduced significantly. This case report suggests that a brain-based model of care may be of value in the chiropractic treatment of migraine and in some non-musculoskeletal conditions. Todd and al. presented a review of literature in 2014 on adverse events due to manual therapies in infants and children.³⁴ There were 31 reported cases of injuries from various healthcare

providers (7 chiropractors, 1 medical practitioner, 1 osteopath, 2 physical therapists and 1 unknown practitioner), 12 presented with serious injuries where an underlying pre-existing pathology was identified. Three deaths were reported from a physical therapist, a craniosacral therapist and an unknown practitioner. The current literature outlined no deaths associated to chiropractic care following manual therapy. This review stresses the importance of thorough history, examination and imaging if necessary to rule-out any potential pathology and congenital anomaly before applying manual therapy. Spinal manipulation remains a non-invasive, low risk therapeutic approach with minimal side effect for pediatric migraine.

Limitations

In order to fully appreciate the long-term benefits of chiropractic care in children presenting with migraine a larger scope of observation should be used ranging from 2 to 5 years instead of 8 months. It is recognized that this a unique case that cannot be generalized to the entire pediatric population affected by migraines. While under 3 months of consistent chiropractic care, the patient experienced improvement in her symptoms. After a 6 week lapse in chiropractic care, symptoms returned and this time the patient was advised to take two types of medication, Elavil and Naproxen. After three weeks of medication without a reduction in symptoms, the patient resumed chiropractic care. Symptoms started to improve again and eventually the patient was able to reduce the medication. The cumulative effect of the medication and chiropractic care in the second scenario may be linked in relieving the symptoms. Whereas initially, chiropractic care was sufficient to reduce the symptoms.

Early identification of possible migraine triggers can greatly influence the course of treatment.⁶ This case report outlines the need for valid research on the effect of spinal manipulation in the treatment of pediatric migraine to provide an evidence-based treatment alternative to the traditional medical treatment. This case also opens the discussion on the benefits cervical manipulations and cranial work may potentially have on sinus drainage and immune system response in autonomic conditions like sinus congestion.

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The importance of clinical examination and collaborative care in the successful chiropractic management of a 31-month-old boy with acquired torticollis: a case report

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ABSTRACT

Introduction: Acquired torticollis in children may present a diagnostic challenge as many different underlying causes are possible, ranging from benign mechanical concerns to serious and potentially life-threatening conditions. **Presenting concerns:** A 31-month old boy presented to a chiropractic clinic with acquired torticollis of possibly traumatic origin. Although it appeared to be a typical muscular torticollis, certain elements of the history and examination findings could point to more serious diagnoses. Thorough intake, physical examination, and interdisciplinary collaboration are paramount for practitioners to help rule out potentially serious conditions before initiating conservative care. **Interventions:** The initial chiropractic evaluation was performed over the course of one week, and included intraprofessional consultation with a pediatric chiropractor. Previous examinations had been performed at a pediatric hospital. Gentle soft tissue and cervical and pelvic joint mobilization, adapted to the patient's age and size, were performed for a total of six visits over a three-week period. **Outcomes:** Upon re-evaluation, complete resolution of the aforementioned symptoms was noted. The results were maintained at follow-up five weeks later. **Conclusion:** Pediatric chiropractic care proved beneficial for this young boy with acquired torticollis. Intraprofessional consultation and collaboration was helpful in the case and should be encouraged.

Keywords: torticollis, diagnosis, mobilization, child, conservative, chiropractic

Introduction

A number of cases describing the conservative treatment of congenital torticollis in children, including with chiropractic care, have been published throughout the years.¹⁻⁴ However, less has been published about acquired torticollis in children and even fewer publications are related to its conservative management.⁵⁻¹⁰ Torticollis is a symptom that can be indicative of a wide range of pathology from benign muscular contractures to severe and possibly life-threatening conditions like tumors or central nervous system (CNS) disorders.¹¹⁻¹³

Torticollis is a relatively common disorder, and although most cases are considered benign and appear to respond well to conservative therapy, a detailed clinical history and complete physical examination are necessary to rule out more serious etiologies before initiating care.¹⁴⁻¹⁶ This report presents the case of a 31-month old boy who presented to a chiropractic clinic with acquired torticollis and illustrates the importance of a thorough evaluation as well as the benefits of intra and inter-professional collaboration and management.

Case presentation

Presenting history

A 31-month old boy of Hispanic origin presented to a chiropractic clinic with neck pain and torticollis, onset approximately 6 weeks prior. No pre-existing neck complaints or conditions were reported. Two weeks prior to the onset of neck pain and torticollis, the child was reported to have fallen while trying to climb a stroller, but the parents did not witness the incident and were therefore unable to describe or confirm it. Initially, the child complained of left knee and ankle pain and was limping. A few days later the torticollis appeared. A week after the incident, he was brought to a pediatric hospital where radiographs of the left knee and ankle were taken and diagnosed as normal. He was diagnosed with an ankle sprain and the parents were told to give ibuprofen for the pain and inflammation. The following week, the child was brought back to the hospital to interrogate the neck pain and torticollis. Blood tests and cervical radiographs were ordered. The radiographs were deemed normal, but the pediatrician noted "signs of inflammation" in the bloodwork and scheduled a cervical computed tomography (CT) scan. The parents were instructed to continue giving ibuprofen as needed. The CT was later cancelled by another pediatrician when additional blood tests revealed normal inflammatory markers.

One month following the hospital visit and 6 weeks after the fall, the child was brought to the chiropractic clinic with persistent limping and a torticollis in left lateral flexion and right rotation. The neck pain was reported more severe at night and the patient had recently begun hitting his forehead with his hands, was fatigued, lethargic and irritable.

Past history

The patient was a healthy 31-month old boy born by C-section. The APGAR score at birth and after 5 minutes was 9/10, he weighed 7.5 lbs and measured 23½" in length. He was diagnosed with deformational plagiocephaly as a baby and his mother reported difficulty breastfeeding at the left breast. Prior to the incident, he was an active boy who enjoyed playing soccer and had good appetite. He slept 7 to 8 hours per night, and was able to sleep in all positions without difficulty. No other health conditions had been diagnosed with the exception of hypersensitivity to mosquito bites (local swelling and itching). He had received the recommended vaccines on schedule, the last reported immunization was done at 18 months of age.

Past medical history

Review of systems revealed an upper respiratory tract infection in the past 6 months, which was treated with antibiotics. The parents mentioned that the symptoms seemed to last for weeks even after the antibiotics, but were fully resolved at the time of consultation.

The parents sought no intervention for the deformational plagiocephaly and breastfeeding difficulties. The pain and inflammation related to the left ankle and knee and neck concerns were treated with ibuprofen as advised by the medical doctor. No other treatments had been rendered. The family history was unremarkable.

Physical examination and significant findings

During the initial visit, only a partial examination was performed due to patient pain and cooperation. General observation revealed the head laterally flexed to the left with a right rotational component. Active range of motion revealed restricted motion in left rotation and flexion. Passive right lateral flexion was painful and passive left rotation induced left rotation of the entire trunk. Patellar reflexes were 3+ bilaterally. Soft tissue palpation revealed hypertonicity in the left sternocleidomastoid (SCM), upper trapezius and right suboccipital muscles. Multiple lymph nodes were palpable along the right posterior cervical lymphatic chain. Joint palpation revealed restrictions in the upper cervical (C0-C1-C2), mid-cervical (C4-C5), upper thoracic (T2-T3) and sacroiliac joints.

Further evaluation was completed during the following vis-

its, and the child became more cooperative as neck mobility improved. After the initial examination, the chiropractor (MHN) decided to consult another chiropractor (DB), who was a more experienced pediatric chiropractor, in order to better prepare the remainder of the examination and to rule out more serious etiologies for torticollis. The clinical history questions, including a more detailed health history, a better description of symptoms and their evolution and more details about the prior hospital visits, were answered by phone between visits 2 and 3. According to the parents' recollection and photographs of the child at various ages, he did not appear to have a pre-existing torticollis. The chiropractor was able, at the third visit, to perform a partial cranial nerve evaluation (III, IV, VI, VII, XI). The child was no longer limping. The plantar reflexes were normal, and deep-tendon reflexes (DTR) were graded normal (2+) for upper and lower extremities (biceps, triceps, stylo-radial (brachioradial), patellar and Achilles), except for the right patellar DTR which was graded as slightly increased at 3+. The hospital radiological report, stating normal studies of the left lower extremity and cervical spine, was obtained at the fourth visit.

Diagnosis

A working diagnosis of muscular torticollis of biomechanical origin was made. The radiological examination performed at the hospital was unremarkable, no neurological signs or symptoms remained, and the cranial nerves that could be evaluated revealed normal function. There was a probable history of a minor trauma (fall), but the absence of an eyewitness made it impossible to ascertain whether the fall precipitated the neck injury. Cervical range of motion, both active and passive, was significantly reduced and painful, especially in left rotation and flexion. No fever was reported, but the child had been given ibuprofen since the incident, which could have masked the fever.

Most observations, at first glance, indicated a mechanical condition. However, the initial injury to the left knee and ankle, the acquired torticollis appearing within a week of that injury, the decreased in cervical range of motion, the presence of inflammatory markers with blood testing, the constitutional symptoms (fatigue, loss of appetite, loss of interest in usual activities, etc.), as well as the history of upper respiratory tract infection in the previous months could indicate signs of more serious types of acquired torticollis, such as inflammatory arthritides, central nervous system (CNS) disorders, or even infectious or neoplastic conditions.^{9-13,16} Although less common, those conditions could not be ruled out completely on the basis of the examinations performed at the chiropractic clinic and the hospital.

Therapeutic intervention

At each visit, cervical active and passive ranges of motion were evaluated, and both joint and soft tissue palpation were performed in order to determine the treatment. After obtaining informed consent from the parents, the first treatment occurred during the first visit in order to relieve some of the symptoms and facilitate further evaluation. The treatment consisted of gentle soft tissue therapy (bilateral SCM and suboccipital muscles, right gluteal muscles) and gentle joint mobilization of the upper cervical area and left sacroiliac joint.

For all visits, soft tissue therapy consisted of light circular massage or pressure points on the affected muscles. Cervical mobilizations involved non-thrust, sustained contacts on cervical structures while positioning the neck in the direction of required correction (either lateral flexion with a skin contact taken over the lateral aspect of the vertebrae or a combination of lateral flexion and slight rotation to the opposite side with a contact near the zygapophyseal joint). Gentle traction or repeated low amplitude passive move-

ments were added in accordance with the child's tolerance and cooperation. The most commonly treated segments were C1-C2 and C4-C5. Pelvic mobilization involved light pressure on the sacrum or ilium, depending on the mobility restriction evaluated by joint palpation.

Follow-up and outcomes

Table 1 describes the child's visits to the chiropractic clinic as well as parent-reported outcomes. The child was seen a total of six times over a three-week period and demonstrated complete resolution of the symptoms. At the seventh visit, two weeks later, the results were maintained and the child was discharged. The parents were instructed to return for follow up in three months or as needed. The child underwent a total of six visits in a three-week period with a good clinical outcome, which was maintained at the follow-up appointment two weeks later (and a follow-up phone call 3 weeks later confirmed results were still maintained). No adverse effects were observed by the chiropractor or reported by the parents.

Table 1. Follow up and outcomes

DATE	TREATMENT	OUTCOME AT NEXT VISIT :
July 21	Initial consultation Partial examination is performed. Treatment #1 STT (suboccipitals, SCM, gluteals) MOB (upper cervical and pelvis)	Subjective increase of 25% left LF Passive F + right LF possible Child able to lie supine Passive left ROT induces body ROT Child slightly more cooperative
July 22	Visit #2. Examination is continued. Treatment #2 STT (suboccipitals, SCM) MOB (upper cervical and pelvis) Advice to stimulate left ROT	Reported improved sleeping Reported less irritability No need for ibuprofen Head almost in neutral position Reduced tension in left trapeze and SCM upon palpation Fewer palpable lymph nodes Passive left ROT 60°, no body ROT
July 28	Visit #3. Examination is continued. Treatment #3 STT (suboccipitals, SCM) MOB (upper cervical, pelvis)	No visible torticollis Active left ROT 45°, passive 65° Child no longer complaining of pain
Aug. 1	Visit #4. Treatment #4 STT (suboccipitals, SCM) MOB (upper cervical)	No pain complaint since last visit Reported improved sleep and increased activity Active left ROT 60°, passive 75°
Aug. 4	Visit #5. Treatment #5 STT (suboccipitals, SCM) MOB (upper cervical)	No pain complaint since last visit Reported resumed playing sports Active neck movements are normal
Aug. 11	Visit #6. Treatment #6 STT (suboccipitals) MOB (upper cervical)	No pain complaint since last visit Reported resumed playing sports Active neck movements are normal
Aug. 25	Visit #7. No treatment. Patient is discharged with advice of consulting on an as-needed basis or follow-up in 3 months time.	

LF : lateral flexion F: flexion ROT: rotation
STT: soft tissue therapy MOB: mobilization

Discussion

Torticollis is a relatively common pediatric condition and the majority of cases are benign in origin.¹⁶ The most common type of torticollis seen in children is congenital muscular torticollis, with a reported incidence ranging from 0.3 to 2%.^{11-12,15,17} Congenital muscular torticollis is typically associated with intrauterine constraint and/or birth trauma, and usually appears within the first month of life, although it may become more apparent in the early childhood years.¹¹⁻¹² Most cases either self-resolve within the first year of life or with conservative treatments consisting of various manual therapy approaches including stretching, postural exercises, massage and postural advice. Some cases may require surgical intervention.¹¹

Contrary to congenital muscular torticollis, acquired torticollis in children can be associated with a myriad of causes.^{7,9-12,14} Among those etiologies, the most common are traumatic, infectious, or CNS-related disorders.^{7,11-12,14,18} Other causes include congenital malformations of the craniocervical structures, as well as tumoral and inflammatory conditions.¹¹⁻¹² It is important to consider these etiologies when evaluating a child with acquired torticollis.

A literature search was done through PubMed to retrieve recent publications on this topic, using the key words torticollis, pseudotumor, dystonia, chiropractic, spinal manipulation, physical therapy, children, congenital, acquired. Papers were selected based on date of publication (2000-2015) and language (French and English), as well as the relevance to conservative care. The search was also widened to related articles and cited references. The literature supports the fact that any acquired torticollis in a child should be considered as traumatic when initially evaluated, as even minor trauma has been reported to induce torticollis.^{11,14,19} The cervical spine of children reacts differently to the deformational forces in acceleration/deceleration injuries.²⁰ This is explained by the increased head to body ratio, the relative ligamentous laxity, the weaker neck muscles as well as the more horizontal orientation of cervical facet joints.^{7,17,19-22} This may also increase the risk for traumatic onset atlantoaxial rotatory fixation (AARF) in children.^{7,19-20,22} It is also important to understand that the absence of a history of trauma does not always rule out a traumatic origin, as children may have difficulty reporting or describing falls or other injuries.^{14,16} Therefore, in the presented case, a traumatic onset should be considered.

Another form of AARF has been reported in the literature and is known as Grisel syndrome, a non-traumatic type of AARF of inflammatory origin, which is closely related to a history of ears-nose-throat (ENT) surgical interventions or infections prior to the development of torticollis. A dif-

ferential diagnosis of Grisel syndrome, or inflammatory AARF, should be included in all cases of non-traumatic torticollis following an ENT or respiratory infection or a surgical intervention in the head and neck areas, as was the case with the patient presented here.^{7,10-11,17,21} Although the precise pathophysiology of Grisel syndrome is not completely understood, it is hypothesized that a hyperemic reaction due to local inflammatory changes following the infection or intervention could cause decalcification near the insertion sites of upper cervical spine ligaments. This, combined with relative ligamentous laxity in children, would predispose for subluxation of the C1-C2 complex. In order to compensate for that instability, contracture of the paravertebral muscles could induce the torticollis.^{7,11,14,16-17, 21-22}

The clinical presentation of AARF, whether traumatic or inflammatory in origin, differs from the typical muscular torticollis. While muscular torticollis often presents as ipsilateral lateral flexion and contralateral rotation of the neck, AARF patients present with ipsilateral rotation of the neck (the chin points to the side of the involved SCM muscle).^{7,11,21} Inability to rotate the head past midline has been reported to be a pathognomonic sign for this condition.^{17,21} Patients presenting with AARF may also have fever or other non-specific symptoms, but do not typically have neurological signs or symptoms.^{14-15,17,19,21} Some cases report increased sedimentation rate and leukocyte count.^{7,21} In the present case, although there were reported inflammatory markers in the blood tests, the head position was not typical of AARF.

In order to rule out congenital malformations and pathological and structural conditions, imaging studies are usually recommended in all children presenting with torticollis. In cases of traumatic onset, radiological evaluation of the cervical spine is the usual initial evaluation, which may be followed by magnetic resonance imaging (MRI) or CT studies.¹⁸ Special consideration should be given to the atlantoaxial alignment, as well as to proper positioning considering the patient's postural asymmetry.^{16,18, 20-21} According to certain authors, in the event of acquired torticollis in children in the absence of trauma, the initial recommended imaging evaluation should be MRI or CT studies.¹⁸⁻¹⁹ However, other studies mention that CT studies, even though more sensitive, are not warranted in the absence of other suggestive symptoms, as they involve significant radiation exposure.²⁰ Such symptoms would include neurological deficits, altered mental status, torticollis, and neck pain.²⁰ In the present case, although a CT study was planned at one point, it was cancelled, reportedly following blood tests that had returned to normal values. However, according to the literature, in this case there was justifiable rationale to perform further diagnostic imaging studies.

Prior to any imaging studies, a complete physical examination and clinical history should be performed. Clinical history should emphasize history of trauma (even minor), history of previous respiratory, or ENT infection or intervention, presence of fever and other non-specific symptoms, presence or absence of pain, review of systems including eyes, head, neck, throat, lungs, and neurological symptoms. Physical examination should include observation and postural evaluation of the entire spine, evaluation of passive and active cervical range of motion, evaluation of cranial nerve function and other neurologic structures, as well as gentle joint and soft tissue palpation.^{12,14,16} Unresolved acquired torticollis lasting more than 7 to 10 days and recurrent episodes of torticollis should be investigated with advanced imaging, as torticollis may be the only presenting symptom of serious conditions such as acute disseminated encephalomyelitis, posterior fossa or spinal cord tumors, AARF, and other disorders of the CNS.^{7,9,14,16} Such conditions could explain the absence of positive outcome after several attempts of conservative management of torticollis, and also cause detrimental delays in proper diagnosis and intervention. One of the major predictive factors of poor prognosis in many cases of acquired torticollis, such as AARF and CNS disorders, is the time interval between the onset of symptoms and the formal diagnosis.^{9,17}

In the present case, there may have been traumatic origin, although there were no eye witnesses to the fall. There were also possible risk factors for AARF, both of traumatic origin (from the fall) or inflammatory origin (previous respiratory infection and antibiotic use, lymphadenopathy, reports of "inflammation signs" in blood tests, unresolved torticollis one month after onset, inability to rotate head past midline). However, the head position was more typical of a muscular torticollis. The main limitation in reporting this case was the difficulty to obtain information from the medical file (blood tests, radiological images, other evaluations). Considering the positive outcome and maintained results, it is likely the child suffered from a typical muscular torticollis, which responded well to conservative treatments. However, it was not possible, based on the available information, to rule out other serious conditions. In the absence of a conclusive diagnosis, any treatment should be initiated gently and with great caution and constant attention to the evolution of the condition. Reported successful conservative management of pediatric torticollis in chiropractic and physical therapy typically include passive mobilizations and stretching exercises, as well as the use of spinal manipulation, especially by chiropractors.¹⁴ Intra-professional collaboration with a practitioner more familiar with pediatric examination and management improved the quality of care in this case, and was likely instrumental in the positive outcomes. However, it proved difficult for the chiropractor to obtain the results

from the medical examinations, which would have facilitated optimal decision-making.

Conclusion

Neck conditions are the second leading reason for patients seeking chiropractic care in Canada and the United States.²³ It is therefore probable that a parent may seek chiropractic care for a child who develops a torticollis. Although manual therapy offered by chiropractors and other health professionals has been shown beneficial in some cases of torticollis, especially the congenital muscular variation, awareness about other types of torticollis is necessary to ensure appropriate management for all patients. In order to ensure that a thorough examination and clinical history are performed prior to initiating treatment, practitioners who are not practiced at performing pediatric evaluations are encouraged to seek advice from colleagues with more expertise with the pediatric population. Although conservative care of pediatric torticollis appears to be effective based on the available publications, more extensive research on a larger scale and with higher methodological quality would be warranted to better document the type of care offered and the outcomes. Finally, it should be expected that clinical information readily be shared between healthcare professionals, as collabora-

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Special Needs Corner

Marfan's Syndrome

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Marfan's Syndrome is an autosomal dominant genetic disorder impacting connective tissue. The mutation alters the manufacture of functional fibrillin-1. A decrease of FN1 leads to an associated decrease of microfibril formation, an increase in growth factors and a decrease in elasticity which results in instability of the tissues impacted. Marfan's affects approximately 1 in 5000 individuals with an estimated 50% transmission to children, however about 25% of all cases are new mutations (i.e. neither parent diagnosed with MFS).¹ Symptomology can include the following observed characteristics: long arms, fingers and legs; generalized tall and thin individuals, pectus excavatum or carinatum, flexible joints, flat feet, crowded teeth or scoliosis. Systems impacted by Marfan's include cardiovascular,

ocular, pulmonary and dural swelling, among others. The two predominant issues for these patients typically are cardiovascular and ocular, with potential for aortic dissection and myopia, glaucoma, early cataract formation and dislocation of the retina and/or the lens. The condition can be fatal early in life, however general life expectancy can be middle to late adulthood.²

Diagnosis of the disorder in adults is done using the Ghent criteria, however in children, it is not a strong diagnostic test and the Kids-Short Marfan survey³ (FIGURE 1) is considered to be more appropriate. Key factors in the diagnosis include family history, dilatation of large arteries (specifically the aorta and pulmonary artery), dislocation of

FIGURE 1. Kid- Short Marfan Score (Kid SMS) (adapted from Mueller et al 2013)

Revised Ghent Criteria (skeletal signs)

Wrist AND/OR thumb sign	Pectus carinatum/excavatum deformity
Hindfoot deformity	Pneumothorax
Dural ectasia	Protrusio acetabuli
Reduced US/LS AND increased	

Required manifestations	Risk category for likelihood of MFS
Dilatation of aortic root + Ectopia lentis	Very high risk
• dilatation of aortic root	High risk (Complete examination of all symptoms of the revised Ghent Criteria is strictly recommended as soon as possible. Patient should see Marfan Syndrome specialists)
+ mitral valve prolapse + tricuspid valve prolapse	
or	
+ dilatation of pulmonary artery or	
+ at least 3 skeletal features of the systemic score of the revised Ghent criteria	
• Ectopia lentis	
+ mitral valve prolapse + tricuspid valve prolapse	
or	
+ dilatation of pulmonary artery	
Family history dilatation of aortic root	Moderate risk (Patient needs to be verified or excluded with further diagnostic procedures other than or echocardiography and clinical examination)

arm/height AND no severe scoliosis	Reduced elbow extension
Scoliosis or thoracolumbar kyphosis	Myopia > 3 diopters
Facial features (dolichocephaly, enophthalmos, downslanting palpebral fissures, malar hypoplasia, retrognathia)	
Skin striae	
Mitral valve prolapse (all types)	

the lens and skeletal anomalies. Diagnosis in a child can be very challenging, as several of the key symptoms are more age dependent and may not be noted in a younger patient. However, should a diagnosis of Marfan's be suspected, genetic testing and follow up with specialists is imperative.⁴

There currently exist no chiropractic care models for Marfan's syndrome; there is a single case report of care to a young adult following a motor vehicle accident with function being returned following whiplash within four visits.⁵ While Marfan's presents a complicated case picture, there are avenues for chiropractic care to assist overall quality of life. Adjusting techniques may need to be altered to accommodate for increased flexibility and hyper mobility. Several of the key skeletal features would be expected to either respond or at least allow for care to have some palliative effect. These would include scoliotic curves, rib restriction and subluxation secondary to sternal deformity and pes planus. Kinesiotaping or similar techniques also could be applied in areas where the skin is not overly fragile to assist with breathing mechanisms, scoliotic correction and normalization of joint motion. Craniosacral therapy may also prove to be of benefit to patients especially around dural swelling, crowding in the mouth or an elevated palate.

Contraindications do exist in this disorder, both absolute and otherwise. A provider needs to be fully aware of the potential or existence of cardiovascular complications, including

but not limited to aortic aneurysms, bronchioectasis and ocular symptoms of lens dislocation and retinal detachment. Technique can be modified to accommodate for a patient's mobility, hyper or restricted, as some joints may develop contracture over time. Care should be provided with full communication with the health team that your patient has developed; if they are beginning their journey, we can assist them in finding literate providers who have expertise with Marfan's Syndrome.

For further information and resources, the patient, parent or provider can visit www.marfan.org.⁶

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PANDAS : A Synopsis

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PANDAS is an abbreviation for Pediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal Infections. This term is used to describe children and adolescents who have OCD (obsessive compulsive disorder) and /or tic disorders that follow or worsen following streptococcal infections.²

Most commonly, OCD and tic symptoms occur gradually over time, but in PANDAS, the symptom onset is unusually abrupt. Often, in PANDAS, the symptoms are noted to occur "out of the blue and overnight" and usually reach full intensity in 24 to 48 hours.³ These OCD symptoms were usually accompanied by other neuropsychiatric symptoms such as anxiety attacks, extreme mood swings, hyperactivity, and difficulty with school subjects.³ These symptoms

usually occurred following a strong stimulant to the immune system such as a viral or bacterial infection.²

Because these symptoms have been noted to increase with intensity following an untreated or partially treated streptococcal infection, the term "pseudo-autoimmune" reaction is used to describe the immune response. This term is used to indicate that the immune system is not really abnormal but causes harmful effects between the anti-strep response and the body's own tissues.² This can be attributed to the ability of the strep bacteria to 'hide' from the immune system for long periods through mimicry of the host tissue. When the immune system finally recognizes the foreign strep, it makes antibodies that not only react to the strep molecules, but with the host tissue that was mimicked. These 'cross-re-

active' antibodies trigger an immune response that attacks the host's own tissues such as brain tissue, which can lead to the OCD and other neuropsychiatric symptoms.²

Diagnosis of PANDAS uses the criteria of: 1. Presence of clinically significant obsessions, compulsions and/or tics; 2. Unusually abrupt onset of symptoms or a relapse-remitting course of symptom severity. This means that during the initial and subsequent occurrences, the OCD symptoms can be plotted to show a worsening immediately following a strep infection.² The OCD symptoms usually decrease significantly and occasionally completely resolve between episodes. Proper testing for streptococcal bacteria is essential to ensure a proper diagnosis.

The best treatment for acute episodes of PANDAS is to eliminate the strep infection causing the symptoms.¹ If a throat culture for strep is positive, a single course of antibiotics will usually get rid of the strep infection and allow PANDAS symptoms to subside. Preventative measures that can be taken are to sterilize or replace things such as toothbrushes or to evaluate family members as to if they are 'strep carriers' so that all sources of the strep infections can be noted and potentially remediated. Prophylactic antibiotics have been shown to be effective to avoid future PAN-

DAS episodes as well.²

To date, there have been no studies directly involving chiropractic care and PANDAS. Because of the relationship the immune system plays in this disease though, it can be postulated that since chiropractic has been shown to increase more natural function of the systems of the body, it may benefit those suffering from PANDAS through a better immune response. This is of course hypothetical and needs to be studied in order to make any direct relation with chiropractic as a treatment for PANDAS, but with the impressive anecdotal evidence from chiropractic and other neuropsychological maladies, it is definitely a worthwhile treatment in conjunction with treatment for active or latent bacterial infection.

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Bracing and exercise-based treatment for idiopathic scoliosis.

Kalichman L, Kendelker L, Bezalel T.

J Bodyw Mov Ther. 2016 Jan;20(1):56-64. doi: 10.1016/j.jbmt.2015.04.007. Epub 2015 Apr 23.

ABSTRACT

BACKGROUND: Various conservative therapies are available for treating adolescent idiopathic scoliosis (AIS), however, the disparities between them and the evidence of their efficacy and effectiveness is still unclear. **OBJECTIVES:** To evaluate the effectiveness of different conservative treatments on AIS. **METHODS:** A literature-based narrative review of the English language medical literature. **RESULTS AND CONCLUSIONS:** The most appropriate treatment for each patient should be chosen individually and based on various parameters. Bracing has been found to be a most effective conservative treatment for AIS. There is limited evidence that specific physical exercises also an effective intervention for AIS. Exercise-based physical therapy, if correctly administered, can prevent a worsening of the curve and may decrease need for bracing. In addition, physical exercises were found to be the only treatment improving respiratory function. Combining bracing with exercise increases treatment efficacy compared with a single treatment. Additional, well-designed and good quality studies are required to assess the effectiveness of different conservative methods in treating AIS.

KEYWORDS: Adolescent idiopathic scoliosis; Bracing; Conservative treatment; Exercises; Review

Current management of pregnancy-related low back pain: a national cross-sectional survey of U.K. physiotherapists.

Bishop A, Holden MA, Ogollah RO, Foster NE; EASE Back Study Team.

Physiotherapy. 2016 Mar;102(1):78-85. doi: 10.1016/j.physio.2015.02.003. Epub 2015 Apr 19.

ABSTRACT

BACKGROUND: Pregnancy-related low back pain (LBP) is very common. Evidence from a systematic review supports the use of exercise and acupuncture, although little is known about the care received by women with pregnancy-related back pain in the U.K. **OBJECTIVE:** To describe current acupuncture and standard care management of pregnancy-related LBP by U.K. physiotherapists. **DESIGN:** Cross-sectional survey of physiotherapists with experience of treating women with pregnancy-related LBP from three professional networks of the Chartered Society of Physiotherapy. **METHODS:** In total, 1093 physiotherapists were mailed a questionnaire. The questionnaire captured respondents' demographic and practice setting information, and experience of managing women with pregnancy-related back pain, and investigated the reported management of pregnancy-related LBP using a patient case vignette of a specific, 'typical' case. **RESULTS:** The overall response rate was 58% (629/1093). Four hundred and ninety-nine physiotherapists had experience of treating women with pregnancy-related LBP and were included in the analysis. Most respondents worked wholly or partly in the U.K. National Health Service (78%). Most respondents reported that they treat patients with pregnancy-related LBP in three to four one-to-one treatment sessions over 3 to 6 weeks. The results show that a range of management strategies are employed for pregnancy-related LBP, and multimodal management is common. The most common reported treatment was home exercises (94%), and 24% of physiotherapists reported that they would use acupuncture with the patient described in the vignette. **CONCLUSIONS:** This study provides the first robust data on the management of pregnancy-related LBP by U.K. physiotherapists. Multimodal management is common, although exercise is the most frequently used treatment for pregnancy-related

KEYWORDS: Acupuncture; Low back pain; Physiotherapy; Pregnancy; Survey

Sick leave and healthcare utilization in women reporting pregnancy related low back pain and/or pelvic girdle pain at 14 months postpartum.

Bergström C, Persson M, Mogren I.

Chiropr Man Therap. 2016 Feb 15;24:7. doi: 10.1186/s12998-016-0088-9. eCollection 2016.

ABSTRACT

BACKGROUND: Pregnancy related low back pain (PLBP) and pelvic girdle pain (PGP) are considered common complications of pregnancy. The long-term consequences for women with persistent PLBP/PGP postpartum are under-investigated. The main objective was to investigate the prevalence, pattern and degree of sick leave as well as healthcare utilization and its perceived effect in women with persistent PLBP/PGP at 12 months postpartum. **METHOD:** This is a follow-up study of a cohort involving of a sample of women, who delivered from January 1(st) 2002 to April 30(th) in 2002 at Umeå University Hospital and Sunderby Hospital, and who reported PLBP/PGP during pregnancy. A total of 639 women were followed-up by a second questionnaire (Q2) at approximately 6 months postpartum. Women with persistent PLBP/PGP at the second questionnaire (N=200) were sent a third questionnaire (Q3) at approximately 12 months postpartum. **RESULTS:** The final study sample consisted of 176 women reporting PLBP/PGP postpartum where N = 34 (19.3 %) reported 'no' pain, N = 115 (65.3 %) 'recurrent' pain, and N = 27 (15.3 %) 'continuous' pain. The vast majority (92.4 %) of women reported that they had neither been on sick leave nor sought any healthcare services (64.1 %) during the past 6 months at Q3. Women with 'continuous' pain at Q3 reported a higher extent of sick leave and healthcare seeking behaviour compared to women with 'recurrent' pain at Q3. Most women with persistent PLBP/PGP had been on sick leave on a full-time basis. The most commonly sought healthcare was physiotherapy, followed by consultation with a medical doctor, acupuncture and chiropractic. **CONCLUSION:** Most women did not report any sick leave or sought any healthcare due to PLBP/PGP the past 6 months at Q3. However, women with 'continuous' PLBP/PGP 14 months postpartum did report a higher prevalence and degree of sick leave and sought healthcare to a higher extent compared to women with 'recurrent' PLBP/PGP at Q3. Women with more pronounced symptoms might constitute a specific subgroup of patients with a less favourable long-term outcome, thus PLBP/PGP needs to be addressed early in pregnancy to reduce both individual suffering and the risk of transition into chronicity.

KEYWORDS: Cohort studies; Female; Healthcare utilization; Pelvic girdle pain; Postpartum period; Pregnancy; Pregnancy complications; Pregnancy related low back pain; Sick leave

Intakes of fish and polyunsaturated fatty acids and mild-to-severe cognitive impairment risks: a dose-response meta-analysis of 21 cohort studies.

Zhang Y, Chen J, Qiu J, Li Y, Wang J, Jiao J.

Am J Clin Nutr. 2016 Feb;103(2):330-40. doi: 10.3945/ajcn.115.124081. Epub 2015 Dec 30.

ABSTRACT

BACKGROUND: The intake of fish and polyunsaturated fatty acids (PUFAs) may benefit cognitive function. However, optimal intake recommendations for protection are unknown. **OBJECTIVE:** We systematically investigated associations between fish and PUFA intake and mild-to-severe cognitive impairment risk. **DESIGN:** Studies that reported risk estimates for mild cognitive impairment (MCI), cognitive decline, dementia, Alzheimer disease (AD), or Parkinson disease (PD) from fish, total PUFAs, total n-3 (ω -3) PUFAs, or at least one n-3 PUFA were included. Study characteristics and outcomes were extracted. The pooled RR was estimated with the use of a random-effects model meta-analysis. A dose-response analysis

was conducted with the use of the 2-stage generalized least-squares trend program. **RESULTS:** We included 21 studies (181,580 participants) with 4438 cases identified during follow-up periods (2.1-21 y). A 1-serving/wk increment of dietary fish was associated with lower risks of dementia (RR: 0.95; 95% CI: 0.90, 0.99; P = 0.042, I(2) = 63.4%) and AD (RR: 0.93; 95% CI: 0.90, 0.95; P = 0.003, I(2) = 74.8%). Pooled RRs of MCI and PD were 0.71 (95% CI: 0.59, 0.82; P = 0.733, I(2) = 0%) and 0.90 (95% CI: 0.80, 0.99; P = 0.221, I(2) = 33.7%), respectively, for an 8-g/d increment of PUFA intake. As an important source of marine n-3 PUFAs, a 0.1-g/d increment of dietary docosahexaenoic acid (DHA) intake was associated with lower risks of dementia (RR: 0.86; 95% CI: 0.76, 0.96; P < 0.001, I(2) = 92.7%) and AD (RR: 0.63; 95% CI: 0.51, 0.76; P < 0.001, I(2) = 94.5%). Significant curvilinear relations between fish consumption and risk of AD and between total PUFAs and risk of MCI (both P-nonlinearity < 0.001) were observed. **CONCLUSIONS:** Fishery products are recommended as dietary sources and are associated with lower risk of cognitive impairment. Marine-derived DHA was associated with lower risk of dementia and AD but without a linear dose-response relation.

KEYWORDS: Alzheimer disease; cognitive impairment; dementia; fish; polyunsaturated fatty acids

What Is Evidence-Based About Myofascial Chains: A Systematic Review.

Wilke J, Krause F, Vogt L, Banzer W.

Arch Phys Med Rehabil. 2016 Mar;97(3):454-61. doi: 10.1016/j.apmr.2015.07.023. Epub 2015 Aug 14.

ABSTRACT

OBJECTIVE: To provide evidence for the existence of 6 myofascial meridians proposed by Myers based on anatomic dissection studies. **DATA SOURCES:** Relevant articles published between 1900 and December 2014 were searched in MEDLINE (PubMed), ScienceDirect, and Google Scholar. **STUDY SELECTION:** Peer-reviewed human anatomic dissection studies reporting morphologic continuity between the muscular constituents of the examined meridians were included. If no study demonstrating a structural connection between 2 muscles was found, articles on general anatomy of the corresponding body region were targeted. **DATA EXTRACTION:** Continuity between 2 muscles was documented if 2 independent investigators agreed that it was reported clearly. Also, 2 independent investigators rated methodologic quality of included studies by means of a validated assessment tool (Quality Appraisal for Cadaveric Studies). **DATA SYNTHESIS:** The literature search identified 6589 articles. Of these, 62 article met the inclusion criteria. The studies reviewed suggest strong evidence for the existence of 3 myofascial meridians: the superficial back line (all 3 transitions verified, based on 14 studies), the back functional line (all 3 transitions verified, based on 8 studies) and the front functional line (both transitions verified, based on 6 studies). Moderate-to-strong evidence is available for parts of the spiral line (5 of 9 verified transitions, based on 21 studies) and the lateral line (2 of 5 verified transitions, based on 10 studies). No evidence exists for the superficial front line (no verified transition, based on 7 studies). **CONCLUSIONS:** The present systematic review suggests that most skeletal muscles of the human body are directly linked by connective tissue. Examining the functional relevance of these myofascial chains is the most urgent task of future research. Strain transmission along meridians would both open a new frontier for the understanding of referred pain and provide a rationale for the development of more holistic treatment approaches.

KEYWORDS: Connective tissue; Fascia; Meridians; Myofascial pain syndromes; Rehabilitation

Cannabis and tobacco exposure in relation to brain morphology: a prospective neuroimaging study in young children.

El Marroun H, Tiemeier H, Franken IHA, et al.

Prenatal Biol Psychiatry. Published online June 16, 2016.

<http://www.biologicalpsychiatryjournal.com/article/S0006-3223%2815%2900692-7/abstract> . Accessed June 23, 2016.

ABSTRACT

Background: Cannabis use during pregnancy has been associated with negative behavioral outcomes and psychopathology in offspring. However, there has been little research evaluating alterations in brain structure as a result of maternal cannabis use. In this prospective study, we investigated the association between prenatal cannabis exposure and brain morphology in young children. **Methods:** We matched 96 children prenatally exposed to tobacco only (without cannabis) with 113 unexposed control subjects on the basis of age and gender and subsequently selected 54 children exposed to prenatal cannabis (mostly combined with tobacco exposure). These children (aged 6 to 8 years) were part of a population-based study in the Netherlands, the Generation R Study, and were followed from pregnancy onward. We assessed brain volumetric measures and cortical thickness in magnetic resonance imaging scans using FreeSurfer. We performed vertexwise analyses in FreeSurfer and linear regression analyses adjusting for relevant covariates using Statistical Package for the Social Sciences. **Results:** Prenatal cannabis exposure was not associated with global brain volumes, such as total brain volume, gray matter volume, or white matter volume. However, prenatal cannabis exposure was associated with differences in cortical thickness: compared with nonexposed control subjects, cannabis-exposed children had thicker frontal cortices. Prenatal tobacco exposure compared with nonexposed control subjects was associated with cortical thinning, primarily in the superior frontal and superior parietal cortices. **Conclusions:** Our findings suggest an association between prenatal cannabis exposure and cortical thickness in children. Further research is needed to explore the causal nature of this association.

Keywords: Brain morphology, Neuroimaging, Pediatric brain development, Population-based study, Prenatal cannabis exposure, Prenatal tobacco exposure

Maternal Exposure to Childhood Trauma Is Associated During Pregnancy With Placental-Fetal Stress Physiology.

Moog NK, Buss C, Entringer S, Shahbaba B, Gillen DL, Hobel CJ, Wadhwa PD.

Biological Psychiatry. May 15, 2016; 79(10):831-839. DOI: <http://dx.doi.org/10.1016/j.biopsych.2015.08.032>

ABSTRACT

Background: The effects of exposure to childhood trauma (CT) may be transmitted across generations; however, the time period(s) and mechanism(s) have yet to be clarified. We address the hypothesis that intergenerational transmission may begin during intrauterine life via the effect of maternal CT exposure on placental-fetal stress physiology, specifically placental corticotropin-releasing hormone (pCRH). **Methods:** The study was conducted in a sociodemographically diverse cohort of 295 pregnant women. CT exposure was assessed using the Childhood Trauma Questionnaire. Placental CRH concentrations were quantified in maternal blood collected serially over the course of gestation. Linear mixed effects and Bayesian piece-wise linear models were employed to test hypothesized relationships. **Results:** Maternal CT exposure (CT+) was significantly associated with pCRH production. Compared with nonexposed women, CT+ was associated with an almost 25% increase in pCRH toward the end of gestation, and the pCRH trajectory of CT+ women exhibited an approximately twofold steeper increase after the pCRH inflection point at 19 weeks gestation. **Conclusions:** To the best of

our knowledge, this finding represents the first report linking maternal CT exposure with placental-fetal stress physiology, thus identifying a potential novel biological pathway of intergenerational transmission that may operate as early as during intrauterine life.

Keywords: Childhood trauma, Developmental programming, Intergenerational transmission, Placental CRH, reconceptual stress, Pregnancy

Effect of childhood maltreatment and brain-derived neurotrophic factor on brain morphology.

van Velzen LS. Schmaal L. Jansen R. Milaneschi Y. Opmeer EM. Elzinga BM. van der Wee NJA. Veltman DJ. Penninx BWJH.

Soc Cogn Affect Neurosci (2016) doi: 10.1093/scan/nsw086 First published online: July 12, 2016.

ABSTRACT

ABSTRACT: Childhood maltreatment (CM) has been associated with altered brain morphology, which may partly be due to a direct impact on neural growth, e.g. through the brain-derived neurotrophic factor (BDNF) pathway. Findings on CM, BDNF and brain volume are inconsistent and have never accounted for the entire BDNF pathway. We examined the effects of CM, BDNF (genotype, gene expression and protein level) and their interactions on hippocampus, amygdala and anterior cingulate cortex (ACC) morphology. Data were collected from patients with depression and/or an anxiety disorder and healthy subjects within the Netherlands Study of Depression and Anxiety (NESDA) (N = 289). CM was assessed using the Childhood Trauma Interview. BDNF Val66Met genotype, gene expression and serum protein levels were determined in blood and T1 MRI scans were acquired at 3T. Regional brain morphology was assessed using FreeSurfer. Covariate-adjusted linear regression analyses were performed. Amygdala volume was lower in maltreated individuals. This was more pronounced in maltreated met-allele carriers. The expected positive relationship between BDNF gene expression and volume of the amygdala is attenuated in maltreated subjects. Finally, decreased cortical thickness of the ACC was identified in maltreated subjects with the val/val genotype. CM was associated with altered brain morphology, partly in interaction with multiple levels of the BDNF pathway. Our results suggest that CM has different effects on brain morphology in met-carriers and val-homozygotes and that CM may disrupt the neuroprotective effect of BDNF.

Keywords: childhood maltreatment, brain-derived neurotrophic factor, BDNF, gene expression, brain structure

Early life stress dampens stress responsiveness in adolescence: Evaluation of neuroendocrine reactivity and coping behavior.

Hsiao YM. Tsai TC. Lin YT. Chen CC. Huang CC. Hsu KS.

Psychoneuroendocrinology. Volume 67 , 86 - 99
DOI: <http://dx.doi.org/10.1016/j.psyneuen.2016.02.004>

ABSTRACT

ABSTRACT: Stressful experiences during early life (ELS) can affect brain development, thereby exerting a profound and long-lasting influence on mental development and psychological health. The stress inoculation hypothesis presupposes that individuals who have early experienced an attenuated form of stressors may gain immunity to its more virulent forms later in life. Increasing evidence demonstrates that ELS may promote the development of subsequent stress resistance, but the mechanisms underlying such adaptive changes are not fully understood. The present study evaluated the impact of fragmented dam-pup interactions by limiting the bedding and nesting material in the cage during postnatal days 2–9, a naturalistic animal model of chronic ELS, on the physiological and behavioral responses to different stressors in adolescent mice and characterized the possible underlying mechanisms. We found that ELS mice showed less social interaction deficits after chronic social defeat stress and acute restraint-tailshock stress-induced impaired long-term potentiation (LTP) and enhanced long-term depression (LTD) in hippocampal CA1 region compared with control mice. The effects of ELS on LTP and LTD were rescued by adrenalectomy. While ELS did not cause alterations in basal emotional behaviors, it significantly enhanced stress coping behaviors in both the tail suspension and the forced swimming tests. ELS mice exhibited a significant decrease in corticosterone response and trafficking of glucocorticoid receptors to the nucleus in response to acute restraint stress. Altogether, our data support the hypothesis that stress inoculation training, via early exposure to manageable stress, may enhance resistance to other unrelated extreme stressors in adolescence.

Keywords: Early life stress, HPA-axis, Stress reactivity, Glucocorticoid receptor, Hippocampus, Mouse

Childhood adversity and psychiatric disorder in young adulthood: An analysis of 107,704 Swedes.

Björkenstam E. Burström B. Vinnerljung B. Kosidou K.

Journal of Psychiatric Research, Volume 77, 67 - 75
DOI: <http://dx.doi.org/10.1016/j.jpsychires.2016.02.018>

ABSTRACT

ABSTRACT: Childhood adversity (CA) is associated with increased risks of psychiatric disorder in young adulthood, but details in this association are less known. We aimed to explore the association of a range of CA indicators with psychiatric disorder in young adulthood, and the impact of age at exposure, disorder type and accumulation of indicators. We capitalized on Sweden's extensive and high-quality registers and analyzed a cohort of all Swedes (N = 107,704) born in Stockholm County 1987–1991. Adversities included familial death, parental substance misuse and psychiatric disorder, parental criminality, parental separation, public assistance reciprocity and residential instability. Age at exposure was categorized as: 0–6.9 years (infancy and early childhood), 7–11.9 years (middle childhood), and 12–14 years (early adolescence). Psychiatric disorders after age 15 were defined from ICD codes through registers. Risks were calculated as Hazard Ratios (HR) with 95% confidence intervals (CI). Results showed that exposure to at least one CA was associated with an increased risk of psychiatric disorder (HR 1.4, 95% CI: 1.3–1.4). Risks were increased for mood, anxiety, and psychotic disorders

and ADHD but not for eating disorders. The risk varied with type of disorder but was similar for all exposure periods. Individuals with multiple (3+) CAs had a two-fold risk of psychiatric disorder (HR 2.0, 95% CI: 1.9—2.1). In conclusion, our findings support the long-term negative impact of CA on mental health, regardless of developmental period of exposure. Given that experience of CA is common, efforts should be put to alleviate the burden of childhood adversities for children, particularly among the most disadvantaged.

Keywords: Childhood adversity, Sensitive period, School performance, Epidemiology, Socioeconomic, Young adulthood

Childhood Trauma and Illicit Drug Use in Adolescence: A Population-Based National Comorbidity Survey Replication—Adolescent Supplement Study.

Carliner H. Keyes KM. McLaughlin KA. Meyers JL. Dunn EC. Martins SS.

Journal of the American Academy of Child and Adolescent Psychiatry. August 2016 Volume 55, Issue 8, Pages 701—708, DOI: <http://dx.doi.org/10.1016/j.jaac.2016.05.010>

ABSTRACT

Objective: Although potentially traumatic events (PTEs) are established risk factors for substance use disorders among adults, little is known about associations with drug use during adolescence, an important developmental stage for drug use prevention. We examined whether childhood PTEs were associated with illicit drug use among a representative sample of US adolescents. **Method:** Data were drawn from the National Comorbidity Survey Replication—Adolescent Supplement (NCS-A), which included adolescents aged 13 to 18 years (N = 9,956). Weighted logistic regression models estimated risk ratios for lifetime use of marijuana, cocaine, nonmedical prescription drugs, other drugs, and multiple drugs. **Results:** Exposure to any PTE before age 11 years was reported by 36% of the sample and was associated with higher risk for use of marijuana (risk ratio [RR] = 1.50), cocaine (RR = 2.78), prescription drugs (RR = 1.80), other drugs (RR = 1.90), and multiple drugs (RR = 1.74). A positive monotonic relationship was observed between number of PTEs and marijuana, other drug, and multiple drug use. Interpersonal violence was associated with all drug use outcomes. Accidents and unspecified events were associated with higher risk for marijuana, cocaine, and prescription drug use. **Conclusion:** Potentially traumatic events in childhood are associated with risk for illicit drug use among US adolescents. These findings add to the literature by illustrating a potentially modifiable health behavior that may be a target for intervention. The results also highlight that adolescents with a trauma history are a high-risk group for illicit drug use and may benefit from trauma-focused prevention efforts that specifically address traumatic memories and coping strategies for dealing with stressful life events.

Keywords: substance use, illicit drugs, adolescents, childhood trauma, maltreatment

The effects of maternal alcohol use disorders on childhood relationships and mental health.

Wolfe, J.D.

Soc Psychiatry Epidemiol (2016):1-10.
doi:10.1007/s00127-016-1264-x

ABSTRACT

Purpose: Despite millions of children living in the turmoil of their parents' active alcoholism or the aftermath of past abuse, research to date has not (1) provided a comprehensive examination of the effects of maternal alcohol use disorders (AUDs) on children's social ties outside of their relationships with parents, or (2) considered whether the number and quality of childhood social ties alter the effects of maternal AUDs on children's mental health. **Method:** Using data from the National Longitudinal Surveys of Youth 1979 Children and Young Adults, analysis examined the influence of maternal AUDs on the number and quality of children's ties with siblings, extended family and family friends, peers, and neighborhood members. The analysis also considered how children's social ties influenced the association between maternal AUDs and children's internalizing and externalizing problems. **Results:** Children of alcoholic mothers had similarly sized networks but more distant relationships with siblings and friends, negative interactions with classmates, and isolating neighborhoods. Controlling for these aspects of children's social ties substantially reduced mental health disparities between children of alcoholic mothers and other children. **Conclusions:** Findings support the view that maternal alcohol use disorders have the potential to damage children's mental health while also setting into motion long-term relationship problems. Future research should examine the networks of children who experience parental AUDs to further clarify the social processes that link parental AUDs to children's mental health.

Keywords: Alcohol use disorder, alcoholism, social ties, childhood, early adolescence, mental health, emotional and behavioral problems, childhood adversity

Commentary

Randomized Trial of Introduction of Allergenic Food in Breast-Fed Infants

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STUDY SUMMARY

This randomized trial, which took place from 2009 to 2012 at a single site in the United Kingdom, evaluated early introduction of multiple potentially allergenic foods in infancy. Exclusively breastfed infants were recruited at age 3 months from a general population (not necessarily at higher risk for allergy). The standard-introduction group was exclusively breast-fed until age 6 months, after which the parents could introduce all foods, including potential allergens.

In the early-introduction group, parents introduced six allergenic foods at age 3 months—beginning with cow's milk, then followed by five other allergenic foods (peanut, egg, sesame, whitefish, and eventually wheat). If the child passed an initial oral challenge without a reaction, the parents were instructed to continue giving the child at least 2 g of each allergenic food twice weekly. If a child reacted to the oral challenge, the parents were instructed to avoid that specific food but continue introduction of the other foods to which the child did not react.

To assess exposure to allergenic foods, the parents completed monthly food diaries for the first 12 months, then quarterly

food diaries until the children were 3 years old. The main outcome of interest was whether the children developed an allergy to one of the six allergenic foods at any time between age 12 and 36 months, as evidenced by a positive response to an oral food challenge.

In general, the trial failed to show a protective effect of early introduction of allergenic foods. For the primary outcome of challenge-proven allergy, 5.6% of the early-introduction group had a food allergy at 36 months, compared with 7.1% of the standard-introduction group (relative risk, 0.80; 95% confidence interval, 0.51-1.25)—a nonsignificant difference.

With respect to specific foods, peanut allergy was documented in 1.2% of children in the early-introduction group vs 2.5% of children in the standard-introduction group—again failing to reach statistical significance. Egg allergy occurred in 3.7% of the early-introduction group compared with 5.4% of the standard-introduction group, also not significantly different. The frequency of allergies to the other foods were all less than 1% and did not differ between the two groups.

The investigators also conducted per-protocol analyses on only the infants who adhered to their group assignment for at least 5 months. Across multiple outcomes, the per-protocol analysis suggested that early introduction of allergenic foods was protective, and these differences did reach statistical significance. Additional analyses looking at the amount of foods consumed generally demonstrated that increasing consumption of allergenic foods correlated with lower risk of developing food allergy at either 12 or 36 months.

The investigators concluded that early introduction of multiple allergenic foods did not protect against development of food allergies in the intention-to-treat analyses, but there appeared to be an inverse dose-response in the secondary analyses.

Viewpoint

Although the results of this trial are negative, as pointed out in an accompanying editorial,¹ several findings are worth noting and suggest that further research should be done.

First, even though the differences in the primary outcome and secondary outcomes in the intention-to-treat analyses were not statistically significant, the allergy frequencies were consistently lower among the early intervention group, regardless of food or outcome evaluated (oral challenge results or immunoglobulin E levels, for example).

Second, this trial was conducted in a general population, whereas the LEAP trial (which had such promising results with early introduction of peanut) enrolled children from high-risk families. It's possible that the hint of protective effect shown in this study might be magnified in a population at greater risk.

Third, the duration of feeding of the allergenic foods was relatively short—from age 6 months to 1 year. The LEAP study suggests that longer administration of allergens may be more likely to lead to favorable outcomes.

Finally, the demonstration of an inverse relationship between the amount of allergen consumed and risk of developing an allergy later on is compelling and suggests that adherence to protocol might have produced better effects.

In summary, although this was a well-designed trial that had negative results, I'm not sure that it yet closes the door on similar investigation in either larger or higher-risk populations.

References

1. Wong GW. Preventing food allergy in infancy—early consumption or avoidance? *N Engl J Med*. 2016 Mar 4. [Epub ahead of print] Medscape Pediatrics © 2016 WebMD, LLC

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