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TABLE OF CONTENTS

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JCCP JOURNAL OF CLINICAL CHIROPRACTIC PEDIATRICS

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Manuscript length will vary with the type of article; in general, manuscripts are expected to be 1,500-3,000 words in length, excluding references, tables and figures. These may vary with the type of article. For case reports and case series, see, "Instructions for Case Reports and Case Series." In general, for manuscripts reporting research studies, the order of components is:

- Introduction: succinctly describe the relevant literature supporting the need for the study.
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- Use footnotes to explain details at bottom of the table (below a horizontal line). Identify using either superscripted lower-case letters or standard footnote symbols (sequence: *,†, ‡, §, ||, ¶, **, ††). Sequence the footnotes in the order text is read–from left to right and then down.
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Abstract
The abstract should be 250 words or fewer. It may be either structured or unstructured. If structured, use the same sections as described below for the components of the report (Introduction, Case Presentation, Intervention and Outcomes, Discussion).

Case Report Components

• **Introduction**: State why this case is unusual or important.

• **Methods**: describe the search engine and key words used to review previously published literature on the subject

• **Case presentation**: Provide a brief summary of the patient's presenting demographics, other relevant characteristics, complaint(s) and related symptomatology.

• **Intervention and outcomes**: Describe the course of treatment, including frequency and duration, and summarize the patient's clinical outcomes, using recognized outcome measures if possible. Include whether informed consent was obtained and if there were any adverse events reported.

• **Discussion**: Succinctly state the important aspects of the case, in terms of its implications for patient care in general, or for specific patient populations or conditions. You may also compare/contrast the case to other cases in the published literature. Be cautious about overstating the importance/implications of your case.

Evidence-based Case Report Instructions

An Evidence-based Case Report (EBCR) is NOT the same as a traditional case report. The EBCR focuses on an answerable clinical question, how it was explored in the search, appraising the results and how it applies to the case, along with the integration of this information with the patient interaction. The final stage in this process is to audit the results.

These are the steps to include:[1,2]

• Brief summary of the chief complaint: 50-100 words

• Briefly describe the clinical case: 250-400 words

• Explain how you developed the clinical question: 200-300 words

• Explain your search for evidence (key words, databases used, number of articles retrieved): 50-100 words

• Evaluate the articles retrieved: critically appraise the evidence for validitiy and relevance: 200-300 words

• Describe how you made your clinical decision by applying these findings to the case, including how you considered and integrated the patient's preferences and values: 250-400 words

• Evaluate your performance: 50-100 words

1. Heneghan C, Badenoch D. *Evidence-based Medicine Toolkit*, 2nd ed. Oxford, UK: Blackwell Publishing, 2006. http://onlinelibrary.wiley.com/doi/10.1002/9780470750605.index/summary (download pdf of "all chapters" for free copy of the publication)

2. Jones-Harris AR. The evidence-based case report: a resource pack for chiropractors. *Clin Chiropr* 2003;6 73-84. (download for free from www.chiro.org/cases/FULL/Evidence-based_Case_Report.pdf)

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https://www-ncbi-nlm-nih-gov.uws.idm.oclc.org/pmc/articles/PMC1126937/pdf/302.pdf

Iran J Pediatr. 2010 Sep; 20(3): 261—268. Evidence Based Medicine in Pediatric Practice: Brief Review
https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3446038/

J Can Chiropr Assoc. 2014 March; 58(1): 6—7. **Evidence-based case reports**
http://pubmedcentralcanada.ca/pmcc/articles/PMC3924510/

3 BMJ. Vol 7, Issue 3, 2002, **Evidence-Based Medicine in Practice: EBM Notebook**
http://ebm.bmj.com/content/7/3/68

Evidence Based Practice: How are we doing?

Evidence Based Practice is the mantra for our practices. We all work very hard to live up to this mantra. It is equally well known that it is virtually impossible to live up to as the evidence is incomplete, at best. This is true for all types of health care, not just chiropractic.

You may notice in this issue that we have decided to give a voice to clinical opinions. After all, the three legs of evidence-based practice are the actual evidence which "proves" a procedure works, practices used by clinicians with years of experience as well as the choice of the patient or parent in the case of pediatrics.

In this issue, we are listening to the voice of clinicians. None are devoid of evidence, but the evidence clearly cannot keep up with innovation in practice. We did not follow the usual stringent protocols that virtually every sentence in a scientific article is referenced. Each clinician has referenced their work to the degree possible. And as a specialist in their field, they do keep up with the evidence. Those high level randomized controlled trials are still very scarce in conservative medicine, not least because they are nearly impossible to do in any type of hands-on therapy. More important in our field is the evidence base that our care is safe, and so far, so good, on that score. We all can look forward to the new pediatricDelphi study being carried out around the world, but based in Australia, which will update everyone on both the safety and effectiveness of chiropractic care for children, along with the evidence for how we should proceed in practice.

We hope that these clinical "pearls" will add tools to your toolbox and help broaden your approach to support your patients. Reading some of the papers of our contributing authors may also help you understand more about how and when collaborative referrals are the next appropriate recommendation to make.

We hope that this issue will bring information and also bring joy to your practices as it offers new lenses with which to look at the diverse clinical problems we face. We do hope that we hear from you, both in letters to tell us your points of view and in future articles of your own research and clinical opinions and case studies.

With warm regards and best wishes during the upcoming joyous season.

The Editors of the Journal of Clinical Chiropractic Pediatrics.

Differentiating the impact of biomechanical forces of labor and delivery vs. the effect of a posterior tongue tie on neonatal and infant feeding dysfunction: a clinical evaluation

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ABSTRACT

A myriad of problems may cause feeding difficulties for the neonatal and infant population. However, the effect of mechanically induced stress, strain, and trauma on the infant and its impact on their ability to feed at breast or by bottle is poorly understood or researched and often goes unrecognized, and therefore, uncategorized. This commentary's aim is to evaluate mechanical trauma associated with the birth process as it contributes to feeding dysfunction as opposed to the current trend to consider most dysfunctional oral motor mechanics the result of a posterior tongue tie. The authors will also briefly review and summarize the relationship between mechanical craniovertebral dysfunction and/or posterior tongue tie as a cause for infant feeding dysfunction based on the best current research. The relevance of this discussion is to promote further observation and research to reach a clear diagnostic understanding of the infant's feeding difficulty as each, on its own, can disrupt oral motor function. Ankyloglossia, and the more recently delineated posterior tongue tie, and their accompanying compensations are hypothesized to result in a concomitant dysfunctional range of motion at the cranio-cervical junction and/or the cervical spine.

Key Words: pediatric chiropractic, cranio-cervical junction, birth trauma, feeding difficulty, breastfeeding difficulty, posterior tongue tie, tethered oral tissues (TOTs).

Introduction

Infants are subject to stress, strain, and trauma associated with the birth process.[1] The most frequent area of impact is to the head and neck with injuries ranging from mild to severe. This trauma has the potential of having a negative impact on range of motion of the head and neck as well as the trunk and extremities. Full and unrestricted range of motion of the head and neck is essential for a normal suck-swallow-breath pattern to occur while feeding at breast or on a bottle. Restriction and discomfort in other parts of the body may also result in difficulty feeding due to the pain that the infant feels when held in a specific feeding position and must be considered in a full evaluation. The authors' goal is to clarify as to whether infant birth stress, strain, or trauma can cause or contribute to infant feeding difficulty. To embark on answering this question, the definition of birth trauma, its prevalence and how it is classified will be explored. Beyond the scope of this paper, it would also be important for investigators to explore how the consequences of birth trauma are expressed or manifested in the neonatal period as well as the associated long-term ramifications.

Birth Trauma

In the United States, birth injuries are estimated to occur in 2.6 percent of births. Superficial and temporary, functional and cosmetic sequelae, disability or even death can result as a consequence of birth-related injuries.[2] The Agency for Healthcare research and quality (AHRQ) in the USA has determined seven categories of birth-related injuries including:

- subdural /intracerebral hemorrhage
- epicranial subaponeurotic hemorrhage
- skeletal injuries
- injuries to spine and spinal cord
- peripheral and cranial nerve injuries
- other types of specified and non-specified birth trauma.[2]

The process of birth, whether spontaneous or assisted, is inherently traumatic for the newborn. Birth-related injuries encompass both mechanical and hypoxic-ischemic events.[2] The exact incidence of mechanical trauma at birth may be underestimated.[2] An incidence of 0.82 percent, prevalence has been estimated at 9.5 per 1000 live births.[3] Less than 2 percent of neonatal deaths result from severe birth trauma.[4] These statistics are based on the most severe outcomes from birth trauma. What are the consequences for the neonate who has less severe (mild or moderate) birth trauma? Neither the percentage of the infant population exposed to minor or moderate mechanical birth injuries, or their outcomes, appear to have been evaluated. Regardless of the reported incidence and prevalence of birth trauma, its true nature and long term sequalae are still poorly understood.[2]

The educational curriculum of pediatric chiropractors who work with the neonate should include the evaluation for the potential sequelae of birth injury or trauma. The evaluation

should begin by obtaining a detailed gestational, labor/delivery and postpartum history. The chiropractor, with a clear understanding of the possibility of in-utero constraint, interventions, or manual manipulations utilized can better understand the mechanism of injury or restriction of the fetus during the pregnancy or birth process. The chiropractor will utilize palpation, as well as neurologic and muscle testing, to formulate a differential diagnosis to help understand the cause of feeding dysfunction. This physical examination is comprised of observations of how the infant moves or does not move during feeding, palpation of the head, neck, mouth, and body, and an elicitation and observation for symmetry of primitive feeding reflexes to determine the status of neurologic function.[5,6] It is important that the chiropractor pursue post graduate education for themselves and in the case of breastfeeding difficulties, consider working collaboratively with a healthcare professional, like an Internationally Board Certified Lactation Consultant (IBCLC). The IBCLC should be able to evaluate the infant's competency at breast or on the bottle including recognizing aspects of dysfunctional breastfeeding mechanics (preferred postures, restricted ranges of motion) and when to make appropriate referrals to chiropractors and osteopaths for adjustment or other manual mobilizations or therapies.[7] Seeking professionals who are trained in performing a functional evaluation of the lips and tongue to rule out connective tissue tethering or "ties" is paramount.[7,8] It is the authors' experience that there may also be other individuals, such as a speech and language pathologist or occupational therapist, who may focus on feeding difficulties. If your dyad's goal is to breastfeed, it is important to communicate and ensure that they have specific expertise, knowledge, and a goal of breastfeeding as a focus on compensations, as the substitute of bottle, cup, and spoon feeding are often immediately implemented.

According to Chaturvedi, et al,[2] the following could be potential red flags that the infant has been subjected to mechanical birth stress, strain, or trauma to the head and neck, thus potentially impacting their ability to suck, swallow, and breathe:

- Malposition (Breech; Transverse)
- Fetal macrosomia
- Maternal diabetes and small pelvis
- Malpresentation (Asynclitic; Occiput posterior)
- Prolonged pushing phase
- Premature rupture of membranes
- Shoulder dystocia or a dystotic labor (60 seconds or more passes between the delivery of the head and torso)
- Augmented or Induced delivery (Pitocin etc.)
- Assisted delivery (Vacuum or forceps, hand assisted, fundal pressures)
- Nuchal cord
- Surgical delivery (Emergency C-section)

The authors have found that birth trauma is typically described in the literature as severe in nature, leaving the clinician or physician to only consider trauma having occurred if there is an obvious deficit or damage as described by Chaturvedi, et al.[2] However, they admit that little is still known about the spectrum of mechanically associated birth trauma and that it is often underestimated. This comment resonates with the authors' clinical experience. In practice, the authors have documented injuries occurring on a spectrum ranging from mild, moderate to severe. As a professional community, chiropractors, or other physicians, recognize that infants can sustain what is considered a minor injury from the birth process (for example, accidental traction on the mandible during manual extraction by either the obstetrician's or midwives' hands or by forceps application) that can impact their ability to feed, grow, and develop optimally. So, how can health care professionals, especially chiropractors and osteopaths (and other practitioners of manual medicine who work with this population), recognize the infants who need attention or care but do not fall in one of the major categories of birth trauma as cited by Chaturvedi, et al?[2]

With the medicalization of birth and the "biomedical tendency to pathologize otherwise normal bodily processes and states,"[9] the introduction of interventions like the use of forceps and vacuum suction has the potential to produce obvious or subtle signs of trauma or strain. Currently, the forceps birth modality is utilized for .05 percent of births and the vacuum extraction birth modality is utilized for 2.5 percent of births in the United States.[10] Moreover, are we able to clinically link biomechanical dysfunction or cranio-vertebral subluxation to different presentations or interventions? First, we must acknowledge that the medical definition of subluxation differs from that of the chiropractic definition.[11-16] In chiropractic journals, we read how the subluxation complex can have an adverse effect on the surrounding nervous system.[17] Nevertheless, the subluxation complex can occur in a variety of presentations and degrees of severity, regardless of the definition, at the atlantooccipital (cranio-cervical) junction. For example, a malpresentation or asynclitic presentation may result in a pressure wound (caput succedaneum) on the top of the head and could result in a cervical spine strain and sprain injury due to a buckling of the spine while under excessive and/or prolonged compressive loading.[18] Additionally, is there an association with the traction or compression of a nuchal cord during delivery with upper cervical subluxation or a suboccipital muscle strain? The prevalence of nuchal cord deliveries is reported to occur between 10-29 percent of deliveries.[19] Studies have shown that traction forces of 8 lbs. usually separate the placenta from the uterus. Tensile strength of umbilical cord indicates that the average load required to break the cord is around 10—14 lbs.[19] In addition, vacuum assisted delivery may also be associated with strain

and sprain of the cervical spine due to the recorded forces applied to effectively complete this procedure. This makes biomechanical sense given the average clinician applied forces used to perform a vacuum delivery range between 10 and 32 pounds-force.[1,20] It is recommended that this procedure is not attempted any longer than 20 minutes nor repeated more than two attempts.[21] This excessive amount of force over a prolonged period can potentially disrupt the normal osteoligamentous integrity of the craniovertebral junction. Moreover, chiropractors and osteopaths have historically attributed the cause of hypothesized cervical spine subluxation complex to abnormal physical stressors being applied to this region.[22-24] Several have published papers discussing how these mechanisms are related to breastfeeding dysfunction.[20,25,26,27]

Research has investigated the amount of force required to buckle (subluxate) the adult and pediatric spine.[18,28] Marchand et. al, found that the osteoligamentous sub-catastrophic load (for an infant cadaver from 0-12 months)[28] is 50 newtons or 12 pounds of tensile traction force.[18,28] Panjabi, et al. found that the average critical load of the osteoligamentous cervical spine, excluding muscular support, in an adult (weighing 70 Kg) is 10.5 N or 2.36 pounds-force.[18] It was discovered that the osteoligamentous spine contributes approximately 20 percent to the minimally needed mechanical stability of the cervical spine, while the rest, nearly 80 percent, is provided by the surrounding neck muscles while under gravity.[18] Keep in mind that we are extrapolating this data to a neonatal spine. The neonatal spine is arguably much less stable under the same amount of pressure, especially when the cervical spine's postural stabilizing muscles are not developed and cannot support the load force during prolonged labor through the cervix or extracted from the birth canal. Observe the normal and acceptable forces applied during common assisted and surgical obstetric procedures in Figure 1, below.

When these figures, ranging from 17–308 newtons are compared with that of the mean chiropractic clinician forces used during a sustained contact on a neonate, 1-20 newtons,[28] there are appreciable differences between the forces used in routine labor and delivery as compared to the forces used in the routine neonatal or infant chiropractic adjustment.

Forces are applied to the neonatal presenting part (head and neck) during an uncomplicated birth as well as when interventions are employed. Excessive forces can be observed during a difficult delivery, including, but not limited to, an asynclitic presentation, manual or assisted deliveries, inefficient or prolonged (> 2 hours) pushing by the mother during dysregulated uterine contractions under the influence of epidural analgesia, shoulder dystocia, occiput posterior presentation, a nuchal chord, or a dystotic uterus. It is also important to note that the pressure gradient will vary due to female body habitus and strength during the pushing phase of a vaginal delivery (with and without neuraxial epidural or anesthesia). The question is, is it possible to calculate the force distributed across the head and neck of a neonate over the period of 1-2 hours during the second stage or pushing phase of labor and delivery or when the obstetrician applies traction or rotation to the head and neck with their hands? Also, consider the force exerted by the vacuum or forceps applied to assist the delivery and potentially save the neonate's life. It seems fair to hypothesize that abnormal forces applied to the head, neck, and associated soft tissues and nerves that are recruited for normal feeding might be injured and might result in feeding dysfunction and craniovertebral pain syndromes.

Unit Comparisons	**Normal Spontaneous Delivery (Grimm/Obrien)**	**Forceps Delivery (non-rotational) (Obrien)**	**Vacuum Device (Mean peak clinician force)**	**Ranges of Traction force with vacuum Ext. @600mmHg**	**Recommended Mean Peak Clinician Force with Adjustment (0-23 mos.) Marchand /Todd)**
Newtons	129	251-309	129-145 (Obrien) 17-99.89 (Grimm)	157-308	Max: 20 Mean: 7.7 <12 wks.: 1-2
Pounds	29	56-69	3.8-32.59 Pop-off > 70	35-69.24	4.4 1.73 .44

Figure .1[1,20,28,29,30]
Pop off: indicates how much pounds-force is required to break the seal between the suction cup and the cranium of the fetus.

Relevant Clinical Anatomy

To better understand this relationship, we need to review the relevant clinical anatomy of the upper cervical spine and feeding system. Eating and swallowing are complex behaviors including both volitional and reflexive activities involving more than 30 nerves and muscles.[31] Subjecting the cranio-cervical junction to abnormal amounts of pressure will result in the subsequent disturbance of the normal alignment between the cranium (C0) and the atlas (C1). The following structures in proximity will be at risk of compromise: the superior cervical ganglion, hypoglossal nerve, genioglossal nerve, and Vagus nerve.[32] How can these structures be negatively impacted by craniovertebral subluxation?

It has been cited in scientific literature that the most common area of cervical spine subluxation occurs at C1, C2 and C3.[33,34] In the authors' clinical experience, this presentation has often been observed. Furthermore, the biomechanical implication must be considered when diagnosing a cause for infant feeding dysfunction, especially when there is evidence of mild to severe mechanical birth stress, strain, or trauma. The architectural concept of "form follows function" is mirrored in the human body by Wolff's Law in degenerative changes.[35] But clinically, we also know that function follows form. When there is a change in cranial or spinal shape, alignment or mobility or range of motion, there will be a subsequent alteration in its function. Normal shape, alignment and mobility or range of motion (form) lend to normal function. Abnormal shape, alignment or mobility or alteration in range of motion is a red flag for abnormal functional performance, and for the purpose of this paper, a red flag for abnormal feeding function.

Overall, sprain, strain, or trauma to this region theoretically hinders the function of the neuro-biomechanical system at the craniovertebral junction. Biomechanical compromise at this level may result in dysregulation of the cranial nerves,[36] as well as restricted (or excessive) joint range of motion and altered muscular activities that are required for safe and efficient feeding, regardless of the perceived severity of the stress, strain, or trauma. In addition to mechanical dysfunction, it is likely and possible that infants with these injuries and noxious stimuli will suffer from craniovertebral myofascial pain syndromes. "In this context, nociceptive fibers that travel with the motor fibers which innervate a particular muscle are possibly involved in pain sensation of the involved muscle and its associated fascia."[32] Nociceptive stimuli plays its own role in elevating sympathetic tone and interfering with relaxed, normal feeding.[7,36,37,38]

Oral Ties

A full discussion of ankyloglossia and tethered oral tissues ("anterior" and "posterior" tongue ties, lip ties and buccal ties) are beyond the scope of this commentary but anatomically refer to ligamentous frena that restrict the range of motion and therefore function of the tongue and lips.[39,40] These restrictions have been considered significant in a number of issues other than feeding including but not limited to reflux,[41-44] airway dysfunction,[42] orthodontic issues[45] and articulation difficulties.[46] To breastfeed successfully, the gape must be wide requiring full range of motion of the temporomandibular joint and the cervical spine and the mandible must be free to hinge (drop) and translate forward (allowing for a "cycling" motion as it comes back up), the lips must create a secure passive seal (with the extended tongue) on the tissue of the breast and the tongue needs to extend, elevate, trough around the nipple and undulate smoothly and rhythmically in a peristaltic wave which is essential for swallowing without risk of aspiration of liquid into the lungs.[47,48]

Restrictive frena are taut ligamentous bands of tissue between the floor of the mouth and the underside of the tongue, between the midline of the lips and gum or the cheek and gum. These restrictive frena alter the mobility of the structures that determine efficient removal of milk from breast or bottle. The ability of the mandible to move freely, separate from the tongue (changing gape width/height) or the tongue moving separately from the mandible (changing the ability of the tongue to elevate the breast tissue to the palate without the mandible following and forcefully compressing the breast tissue) is affected by a tongue tie. Efficiency of milk transfer will be affected by the quality of the seal of the lips on the breast tissue. This seal is poor when there is an inability to passively flange the lips due to the presence of taut connective tissue bands between the gum and cheek (buccal ties) or midline frena between the upper or lower lip and gum. When the seal is insecure, there is a greater risk of aerophagia (swallowing air), leakage, and poor milk transfer from the positive pressure of the full breast to the negative pressure of the oral cavity.[8,47,48] An infant may compensate by recruiting the orbicularis oris to hold the breast tissue resulting in "milk blisters" in the midline of the lip or cobbling or cross striations of the entire lip occurring either on one or both of the lips.

A tongue tie may present anteriorly at the tip of the tongue causing a characteristic indentation or crease in the midline or have a more posterior presentation and can be visible upon elevating the tongue or be hidden, embedded in the mucosa and may cause a central indentation or persistent retraction of the tongue. Based on its point of attachment and its "flexibility" (which is dictated by composition of elastin and collagen), both anterior and posterior tongue ties may affect the tongue's ability to extend, elevate or lateralize and often causes a "humping" or retraction of the tongue which will decrease, at best, the efficiency of transfer of milk and at worst, decrease the patency of the pharyngeal aperture (potentially causing airway obstruction) or create

poor channeling of milk (increasing the risk of choking/aspirating).[40]

Any or all interference with latch and efficient milk transfer (using less energy/calories to extract the milk than the energy/calories contained in the milk itself) will result in a sympathetic response in the infant. Due to their efficient neural plasticity, a quick adaptation and reorganization from their "preprogrammed" neurology that guides feeding, the infant may develop a compensatory sequence of motions. This can become evident when the infant recruits accessory muscles that in turn risk compromising other muscles responsible for diverse physiologic functions. This cascade can affect everything from swallowing and breathing, to posture and joint range of motion, increased flexor tone, retained fetal posture, a head tilt with or without rotation or other preferential postures. These compensatory postures or compensatory muscle actions are often the root of the segmental dysfunction or subluxation that can be addressed by the chiropractor or osteopath. The importance of recognizing both issues lie in the fact that treating one or the other exclusively may not have an optimal outcome.

If the subluxation is a result of the aberrant oral motor activity dictated by the presence of tethered oral tissues like a tongue tie, then one would be repeatedly addressing the subluxation secondary to compensatory muscular recruitment without resolution until, perhaps the infant is no longer feeding at breast or on bottle.

On the other hand, surgically intervening by releasing the taut frena will not necessarily result in improved oral motor function if the subluxation (segmental motor dysfunction) is interfering with the infant's ability to gape widely or extend at the base of the cranium.

The authors would also like to point out that there are certainly other unexplored areas of consideration including the epigenetic effects of ankyloglossia or other tethered oral tissues, in-utero constraint resulting in fascial restriction and potential segmental dysfunction on the fetus as well as the epigenetic effects of birth trauma and dysfunctional oral motor function on the neonate.

It is critical for inter-collegial discourse and professional development of an inclusive evaluation and differential diagnosis so that the treatment planning and goals are prioritized and collaboratively delivered to have the best outcome for the dyad.[7,36,37,38]

Discussion

Over the past five years, the lead author has worked in a breastfeeding medical office and has cared for more than 2,000 neonates and infants in a multidisciplinary and collaborative setting with IBCLC's, nurses and a medical physician. A birth history was obtained from the parents of each infant. Every one of these parents reported a chief complaint associated with structural issues in the head and neck while also having feeding difficulties. Some had been previously diagnosed with an anterior tongue tie, or posterior tongue tie (or other oral frena restricting the normal action of the lips, tongue or cheeks), as well as some having no apparent tongue or other oral frena restricting oral motor function, yet were experiencing oral motor dysfunction.[39,48] Some had undergone a surgical procedure that released the tethering oral tissues but had no pre or post-surgical manual or chiropractic care and experienced no improvement in breastfeeding.

There is much debate on the prevalence and incidence of posterior tongue tie.[49-52] Based on published observations, the current research and the author's clinical observation, there is an unexplored incidence of mild to severe mechanical birth stress, strain, and trauma in the infant population which might explain the rise in infant feeding difficulties that can occur with or without other comorbidities like a posterior tongue tie or other tethered oral tissues.[20,38]

Based on these observations, it is critical to differentially diagnose the reasons for breast feeding difficulties. A biomechanical injury to the head and neck should not be confused with a "posterior tongue tie" or any other anatomical restriction of the oral structures by ligamentous structure.[20,26,53,54,55,56] There is mounting research[40] and clinical evidence that a significant cause of infant feeding dysfunction can occur due to abnormal forces during labor and delivery on the cranium, hyoid bone, and cervical spine, which can often masquerade as a "posterior tongue tie." The hyoid, for example, when restricted by a nuchal cord, can result in a change in the muscular action of the muscles of the floor of the mouth, the neck and the tongue, as well as the muscles that influence the range of motion at the craniocervical junction.[36] The position and range of extension and elevation of the tongue can be reduced as a result of hyoid displacement. This may give the appearance of a tongue tie but is actually a "faux tie" (as coined by Hazelbaker)[57] and the hyoid should be mobilized before assessing tongue function. Failure to assess and address biomechanical dysfunction can lead to an unnecessary or premature surgery and/or poor surgical outcome, resulting in continued feeding dysfunction.[7,36,37,38]

Many health professionals interfacing with these infants are not trained how to assess breastfeeding mechanics or appropriately refer infants with biomechanical dysfunction contributing to breastfeeding dysfunction for treatment. Chiropractic physicians are well positioned to educate the public and professional communities on this topic. A literature review of chiropractic care for breastfeeding newborns was performed in 2015 which briefly touched on

the topic of birth trauma as a contributing factor.[25] Further investigation and a review of the literature on the topic of birth trauma and associated infant feeding dysfunction is warranted.

Just as there is much debate on the prevalence and incidence of posterior tongue tie and safety and necessity for their surgical release,[40,58,59] there also seems to be a large debate on the necessity, safety, and efficacy of infant chiropractic care.[60,61] (One must remember to compare the forces of labor and delivery as well as the interventions employed to the forces used during the infant chiropractic adjustment which is recommended to be performed with 1/10th of the force used for adult manipulation.)[28]

Furthermore, the adjustment is an appropriate therapeutic intervention to treat an upper cervical strain and sprain injury or subluxation complex.[28,30,60,61] It is apparent that without adequate care and early intervention, these upper cervical "conditions" resulting from postural loading mechanisms are not self-limiting and continue as an adaptation from normal. These spinal and cranial structural adaptations will potentially perpetuate into other sequela thus leading to less than optimal neural and structural function and in the infant, potentially compromise their development.[62-66]

Mechanical stress and strain to the cranium and vertebral column needs to be considered in the top differential diagnosis when considering the cause of feeding dysfunction or when considering a tethered oral tissue as the primary diagnosis not only for the preservation of the breastfeeding relationship but in consideration of the infants overall development.

Conclusion

Based on the current research and the authors' clinical experience, there seems to be higher than reported prevalence and incidence of mild to moderate and moderate to severe mechanical birth stress, strain, and trauma in the infant population. A potential rise in birth stress, strain, and trauma, with or without ankyloglossia, may be another explanation of the increasing number of infant feeding difficulties and should not be misdiagnosed or mistreated as a posterior tongue tie. This misdiagnosis or the failure to recognize a concomitant situation could result in a less than optimal outcome. Due to the paucity of research available on both topics, the question remains unanswered as to whether there is a current exponentially growing number of children with the structural occurrence of posterior tongue tie or if there is a failure to recognize mild to moderate birth trauma sequelae like (breast) feeding dysfunction. Without reaching a collaborative consensus, there is the risk of normalizing anatomical variants or mechanically induced dysfunction interfering with breastfeeding instead of creating an avenue of support for the breastfeeding dyad.

Further research and investigation on the mechanism of mild to moderate mechanical birth stress, strain, and trauma's (as well as the posterior tongue tie's) effect on infant feeding function and overall development is warranted.

References:

1. Grimm MJ. Forces Involved with Labor and Delivery–A Biomechanical Perspective. *Annals of Biomedical Engineering*. 2021;49(8):1819-1835. doi:10.1007/s10439-020-02718-3.

2. Chaturvedi A, Chaturvedi A, Stanescu AL, Blickman JG, Meyers SP. Mechanical birth-related trauma to the neonate: An imaging perspective. *Insights into Imaging*. 2018;9(1):103-118. doi:10.1007/s13244-017-0586-x.

3. Rabelo NN, Matushita H, Cardeal DD. Traumatic brain lesions in newborns. *Arquivos de Neuro-Psiquiatria*. 2017;75(3):180-188. doi:10.1590/0004-282x20170016.

4. Reichard R. Birth Injury of the Cranium and Central Nervous System. *Brain Pathology*. 2008;18(4):565-570. doi:10.1111/j.1750-3639.2008.00205.x.

5. Naqvi U, Sherman A. Muscle Strength Grading. [Updated 2021 Sep 2]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2022 Jan. Available from: https://www.ncbi.nlm.nih.gov/books/NBK436008/.

6. Futagi Y, Toribe Y, Suzuki Y. "The Grasp Reflex and Moro Reflex in Infants: Hierarchy of Primitive Reflex Responses." *International Journal of Pediatrics*, vol. 2012, 2012, pp. 1—10. https://doi.org/10.1155/2012/191562.

7. Tow, J, Vallone S. Development of an integrative relationship in the care of the breastfeeding newborn: Lactation consultant and chiropractor. *J Clin Chiropr Pediatr.* 2009 June; 10(1):626-632.

8. Elad D, Kozlovsky P, Blum O, Laine AF, Po MJ, Botzer E, Dollberg S, Zelicovich M, Ben Sira L. Biomechanics of milk extraction during breast-feeding. *Proc Natl Acad Sci USA*. 2014 Apr 8;111(14):5230-5. doi: 10.1073/pnas.1319798111. Epub 2014 Mar 24. PMID: 24706845; PMCID: PMC3986202.

9. Inhorn MC. Defining Women's Health: A Dozen Messages from More than 150 Ethnographies. *Medical Anthropology Quarterly*. 2006;20(3):345-378. doi:10.1525/maq.2006.20.3.345.

10. Michas, F. *Forceps or vacuum extraction births U.S. 1990-2020*. Statista. 2022, May 17. Retrieved September 19, 2022, from https://www.statista.com/statistics/276067/us-births-delivered-by-forceps-or-vacuum-extraction/.

11. Johnson C. Use of the term subluxation in publications during the formative years of the chiropractic profession. *J Chiropr Humanit*. 2011;18(1):1-9. doi:10.1016/j.echu.2011.10.004.

12. Bogduk N, Mercer S. Biomechanics of the cervical spine. I: Normal kinematics. *Clinical biomechanics*. Nov 2000;15(9):633-648.

13. Ishii K, Chiba K, Maruiwa H, Nakamura M, Matsumoto M, Toyama Y. Pathognomonic radiological signs for predicting prognosis in patients with chronic atlantoaxial rotatory fixation. *J Neurosurg Spine*. Nov 2006;5(5):385-391.

14. Fielding JW, Hawkins RJ. Atlanto-axial rotatory fixation. (Fixed rotatory subluxation of the atlanto-axial joint). *The Journal of bone and joint surgery*. American volume. Jan 1977;59(1):37-44.

15. Warner WC, Hedequist DJ. Cervical Spine injuries in Children. In: Beaty J, Kasser J, eds. *Fractures in Children*. Vol 1. 8th ed. Philadelphia, PA: Lippincott Williams & Wilkins; 2015: 845-898.

16. Hedequist D, Hresko T. Proctor M. Modern cervical spine instrumentation in children. *Spine*. Feb 15 2008;33(4):379-383.

17. Marcon AR, Murdoch B, Caulfield T. The "subluxation" issue: an analysis of chiropractic clinic websites. *Arch Physiother.* 2019;9:11. Published 2019 Nov 13. doi:10.1186/s40945-019-0064-5.

18. Panjabi MM, Cholewicki J, Nibu K, Grauer J, Babat LB, Dvorak J. Critical load of the human cervical spine: an in vitro experimental study. *Clinical Biomechanics*. 1998;13(1):11-17. doi:10.1016/s0268-0033(97)00057-0.

19. Peesay M. Nuchal cord and its implications. *Matern Health Neonatol Perinatol*. 2017 Dec 6;3:28. doi: 10.1186/s40748-017-0068-7. PMID: 29234502; PMCID: PMC5719938.

20. Miller JE, Miller L, Sulesund AK, Yevtushenko A. Contribution of Chiropractic Therapy to Resolving Suboptimal Breastfeeding: A Case Series of 114 Infants. *Journal of Manipulative & Physiological Therapeutics*. 2009;32(8):670-674. doi:10.1016/j.jmpt.2009.08.023.

21. Putta LV, Spencer JP. Assisted vaginal delivery using the vacuum extractor. *Am Fam Physician*. 2000;62(6):1316-1320.

22. Frymann VM, Carney RE, Springall P. Effect of osteopathic medical management on neurologic development in children. *J Am Osteopath Assoc*. 1992;92(6):729-744.

23. Brurberg KG, Myrhaug HT, Reinar LM. Diagnostics and Treatment of Infants Suspected with Kinematic Imbalance Due to Suboccipital Strain (KISS). Knowledge Centre for the Health Services at The Norwegian Institute of Public Health (NIPH), Oslo, Norway; 2009. PMID: 29320073.

24. Biedermann, H. (1992). Kinematic imbalances due to suboccipital strain in newborns. *J Manual Med*, 6, 151-156.

25. Alcantara J, Alcantara JD, Alcantara J. The Chiropractic Care of Infants with Breastfeeding Difficulties. *EXPLORE*. 2015;11(6):468-474. doi:10.1016/j.explore.2015.08.005.

26. Hawk C, Minkalis A, Webb C, Hogan O, Vallone S. Manual Interventions for Musculoskeletal Factors in Infants With Suboptimal Breastfeeding: A Scoping Review. *Journal of Evidence-based Integrative Medicine*. 2018;23. doi:10.1177/2515690X18816971.

27. Hom S, Shikada K. Resolution of allergic colitis, colic, plagiocephaly, and breastfeeding challenges following chiropractic in an infant with birth trauma: Case study [case report]. *J Pediatr Matern & Fam Health - Chiropr*. 2021 Jul;2021():53-59. ICLID: 27003.

28. Marchand AM. A Proposed Model With Possible Implications for Safety and Technique Adaptations for Chiropractic Spinal Manipulative Therapy for Infants and Children. *Journal of Manipulative and Physiological Therapeutics*. 2015;38(9):713-726. doi:10.1016/j.jmpt.2013.05.015.

29. O'Brien SM, Winter C, Burden CA, Boulvain M, Draycott TJ, Crofts JF. Pressure and traction on a model fetal head and neck associated with the use of forceps, Kiwi™ ventouse and the BD Odon Device™ in operative vaginal birth: a simulation study. *BJOG*. 2017;124 Suppl 4(Suppl 4):19-25. doi:10.1111/1471-0528.14760.

30. Todd AJ, Carroll MT, Mitchell EKL. Forces of Commonly Used Chiropractic Techniques for Children: A Review of the Literature. *Journal of Manipulative and Physiological Therapeutics*. 2016;39(6):401-410. doi:10.1016/j.jmpt.2016.05.006.

31. Golding S. Normal and abnormal swallowing. Imaging in diagnosis and therapy (2nd edn). Edited by B Jones, pp. xviii + 287, 2003 (Springer-Verlag, New York, NY), £122.00 ISBN 0-387-95194-6. *The British Journal of Radiology*. 2004;77(917):458-458. doi:10.1259/bjr.77.917.770458b.

32. Sutcliffe P, Lasrado S. Anatomy, Head and Neck, Deep Cervical Neck Fascia. National Library of Medicine: National Center for Biotechnology Information. StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2020. Accessed December 6, 2020. https://www.ncbi.nlm.nih.gov/books/NBK541091/.

33. Ahn AR, Rah UW, Woo JE, Park S, Kim S, Yim SY. Craniovertebral Junction Abnormalities in Surgical Patients With Congenital Muscular Torticollis. *J Craniofac Surg*. 2018;29(3):e327-e331. doi:10.1097/SCS.0000000000004403.

34. Slate RK, Posnick JC, Armstrong DC, Buncic JR. Cervical Spine Subluxation Associated with Congenital Muscular Torticollis and Craniofacial Asymmetry. *Plastic and Reconstructive Surgery*. 1993;91(7):1187-1195. doi:10.1097/00006534-199306000-00001.

35. Brand, Richard A.1, a. Biographical Sketch: Julius Wolff, 1836-1902. *Clinical Orthopaedics and Related Research*: April 2010 - Volume 468 - Issue 4 - p 1047-1049 doi: 10.1007/s11999-010-1258-z.

36. Vallone S. Evaluation and treatment of breastfeeding difficulties associated with cervicocranial dysfunction: a chiropractic perspective. *J. of Clinical Chiropr Pediatr.* 2016.15(3):1301-1305.

37. Vallone, S., Carnegie-Hargreaves F. The Infant with Dysfunctional Feeding Patterns- The Chiropractic Assessment. *J. of Clinical Chiropr Pediatr*. 2016 May; 15(2):1230-1235.

38. Vallone, Sharon. (2004). Chiropractic evaluation and treatment of musculoskeletal dysfunction in infants demonstrating difficulty breastfeeding. *J. of Clinical Chiropr Pediatr*. 2004 Dec; 6 (1): 349—366.

39. Becker S, Mendez MD. Ankyloglossia. [Updated 2022 Mar 15]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2022 Jan-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK482295/.

40. Mills N, Keough N, Geddes DT, Pransky SM, Mirjalili SA. Defining the anatomy of the neonatal lingual frenulum. *Clin Anat*. 2019;32(6):824-835. doi: 10.1002/ca.23410.

41. Siegel SA. Aerophagia Induced Reflux in Breastfeeding Infants With Ankyloglossia and Shortened Maxillary Labial Frenula (Tongue and Lip Tie). *Int J Clin Pediatr*. 2016;5(1):6-8. doi: http://dx.doi.org/10.14740/ijcp246w.

42. Kotlow, L. Tethered oral tissues as a differential diagnostic tool in infants and toddlers presenting with obstructive sleep apnoea and air induced reflux. *Australasian Medical Journal*. 2019;12(5):131-137.

43. Hill RR, Pados BF. Gastrointestinal Symptom Improvement for Infants Following Tongue-Tie Correction. *Clinical Pediatrics*. 2022;0(0). doi:10.1177/00099228221117459.

44. Hand P, Olivi G, Lajolo C, et al. Short lingual frenum in infants, children and adolescents. Part 1: breastfeeding and gastroesophageal reflux disease improvement after tethered oral tissues release. *Eur J Paediatr Dent*. 2020;21(4):309-317.

45. Yoon AJ, Zaghi S, Ha S, Law CS, Guilleminault C, Liu SY. Ankyloglossia as a risk factor for maxillary hypoplasia and soft palate elongation: A functional - morphological study. *Orthod Craniofac Res*. 2017 Nov;20(4):237-244. doi: 10.1111/ocr.12206. Epub 2017 Oct 10. PMID: 28994495.

46. Baxter R, Hughes L. Speech and Feeding Improvements in Children After Posterior Tongue-Tie Release: A Case Series. *International Journal of Clinical Pediatrics* 7 (2018): 29-35.

47. Geddes DT, Sakalidis VS. Ultrasound Imaging of Breastfeeding--A Window to the Inside: Methodology, Normal Appearances, and Application. *J Hum Lact*. 2016 May;32(2):340-9. doi: 10.1177/0890334415626152. Epub 2016 Feb 29. PMID: 26928319.

48. LeFort Y, Evans A, Livingstone V, Douglas P, Dahlquist N, Donnelly B, Leeper K, Harley E, Lappin S. Academy of Breastfeeding Medicine Position Statement on Ankyloglossia in Breastfeeding Dyads. *Breastfeed Med*. 2021 Apr;16(4):278-281. doi: 10.1089/bfm.2021.29179.ylf. PMID: 33852342.

49. Segal LM, Stephenson R, Dawes M, Feldman P. Prevalence, diagnosis, and treatment of ankyloglossia: methodologic review. *Can Fam Physician*. 2007;53(6):1027-1033.

50. Fraser L, Benzie S, Montgomery J. Posterior tongue tie and lip tie: a lucrative private industry where the evidence is uncertain. *BMJ*. Published online November 26, 2020:m3928. doi:10.1136/bmj.m3928.

51. Solis-Pazmino P, Kim GS, Lincango-Naranjo E, Prokop L, Ponce OJ, Truong MT. Major complications after tongue-tie release: A case report and systematic review. *International Journal of Pediatric Otorhinolaryngology*. 2020;138(138):110356. doi:10.1016/j.ijporl.2020.110356.

52. Srinivasan A, Al Khoury A, Puzhko S, et al. Frenotomy in Infants with Tongue-Tie and Breastfeeding Problems. *Journal of Human Lactation*. 2018;35(4):706-712. doi:10.1177/0890334418816973.

53. Ghaheri BA, Lincoln D, Mai TNT, Mace JC. Objective Improvement After Frenotomy for Posterior Tongue-Tie: A Prospective Randomized Trial. *Otolaryngology—Head and Neck Surgery*. 2021;166(5):976-984. doi:10.1177/01945998211039784.

54. Miller J, Beharie MC, Taylor AM, Simmenes EB, Way S. Parent Reports of Exclusive Breastfeeding After Attending a Combined Midwifery and Chiropractic Feeding Clinic in the United Kingdom: A Cross-Sectional Service Evaluation. *J Evid Based Complementary Altern Med*. 2016 Apr;21(2):85-91. doi: 10.1177/2156587215625399. Epub 2016 Jan 13. PMID: 26763046; PMCID: PMC4768400.

55. Holleman AC, Nee J, Knaap SF. Chiropractic management of breast-feeding difficulties: a case report. *Journal of Chiropractic Medicine*. 2011; 10(3), 199-203.

56. Holtrop DP. Resolution of suckling intolerance in a 6-month-old chiropractic patient. *Journal of Manipulative and Physiological Therapeutics*. 2000;23(9): 615-618.

57. Hazelbaker A. *Tongue-tie morphogenesis, impact, assessment and treatment*. Columbus, Ohio: Aiden and Eva Press; 2010.

58. O'Callahan C, Macary S, Clemente S. The effects of office-based frenotomy for anterior and posterior ankyloglossia on breastfeeding. *Int J Pediatr Otorhinolaryngol* 2013;77:827—832.

59. Ghaheri BA, Cole M, Fausel SC, Chuop M, Mace JC. Breastfeeding improvement following tongue-tie and lip-tie release: a prospective cohort study. *Laryngoscope*. 2017;127(5):1217-1223. doi:10.1002/lary.26306.

60. Caloway C, Hersh CJ, Baars R, Sally S, Diercks G, Hartnick CJ. Association of Feeding Evaluation With Frenotomy Rates in Infants With Breastfeeding Difficulties. *JAMA Otolaryngol Head Neck Surg*. 2019;145(9):817—822. doi:10.1001/jamaoto.2019.1696.

61. Todd AJ, Carroll MT, Robinson A, Mitchell EKL. Adverse Events Due to Chiropractic and Other Manual Therapies for Infants and Children: A Review of the Literature. *Journal of Manipulative and Physiological Therapeutics*. 2015;38(9):699-712. doi:10.1016/j.jmpt.2014.09.008.

62. Driehuis F, Hoogeboom TJ, Nijhuis-van der Sanden MWG, de Bie RA, Staal JB. Spinal manual therapy in infants, children and adolescents: A systematic review and meta-analysis on treatment indication, technique and outcomes. *PLoS One*. 2019 Jun 25;14(6):e0218940. doi: 10.1371/journal.pone.0218940. PMID: 31237917.

63. Harrison, DE, Cailliet, R, Harrison, DD, Troyanovich, SJ, & Harrison, SO (1999). A review of Biomechanics of the central nervous system–part III: Spinal cord stresses from postural loads and their neurologic effects. *Journal of Manipulative and Physiological Therapeutics*, 22(6), 399—410. https://doi.org/10.1016/s0161-4754(99)70086-2.

64. Maningat AL, Sunil Munakomi. Neuroanatomy, Superior Cervical Ganglion. Nih.gov. Published June 22, 2019. Accessed December 21, 2019. https://www.ncbi.nlm.nih.gov/books/NBK544331/.

65. Bordoni B, Morabito B, Mitrano R, Simonelli M, Toccafondi A. The Anatomical Relationships of the Tongue with the Body System. *Cureus*. 2018;12. doi:10.7759/cureus.3695.

66. Koch LE, Koch H, Graumann-Brunt S, Stolle D, Ramirez JM, Saternus KS. Heart rate changes in response to mild mechanical irritation of the high cervical spinal cord region in infants. *Forensic Science International*. 2002;128(3):168-176. doi:10.1016/s0379-0738(02)00196-2.

Manual therapy by chiropractors for infants with musculoskeletal-related suboptimal infant breastfeeding: a pilot study

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ABSTRACT

Objective: to assess one-week outcomes of manual therapy by chiropractors for infants with musculoskeletal dysfunction and suboptimal infant breastfeeding (SIB), using the MIBAQ (Musculoskeletal Infant Breastfeeding Questionnaire). **Methods:** This was a descriptive cohort study. Volunteer chiropractors who frequently treat infants with musculoskeletal dysfunctions enrolled eligible infants within a two-month study period. Mothers of infants ≤ 3 months currently or recently breastfeeding presenting for care in the participating office were eligible. Mothers who declined to participate were excluded. The primary outcome was MIBAQ change from pre-treatment to one week later. A secondary measure was the Patient's Global Impression of Change. **Results:** A total of 72 participants from six chiropractic offices completed the pre-survey; 35 (49%) completed both pre- and post-survey. The MIBAQ scores improved highly significantly ($p < .000$) from pre- to post-treatment and were significantly correlated with the PGIC (Pearson correlation=.408; p=.021). **Conclusions:** These results demonstrate that the MIBAQ appears to be clinically responsive to changes in SIB-related symptoms and could facilitate larger practice-based research studies of infants with musculoskeletal dysfunction contributing to SIB. In this study, significant clinical change was reported by mothers of infants with SIB after one week of chiropractic manual therapy.

Key Words: suboptimal breastfeeding, outcome assessment tool, chiropractic.

Introduction

Exclusive breastfeeding for the first six months of life has been recommended by authorities worldwide.[1,2] The World Health Organization further states that breastfeeding in combination with age-appropriate foods should continue until the child is at least two years old.[2] Breastfeeding that does not meet the minimum medical recommendation is termed suboptimal infant breastfeeding (SIB),[3] and the current prevalence of SIB is reported at 44% with SIB correlated to 16% of child and 78% of maternal excess deaths annually.[2,4]

Although the numbers are daunting, women continue to seek and utilize alternative forms of providing nutrients to their children with a list of reasons as varied as the mothers themselves.[5,6] On the community level and policy level, physicians, nurses, and chiropractors along with public health agencies such as the US Preventative Services Task Force (USPSTF) continue to encourage breastfeeding.[7,8]

Biomechanically, authorities in the nursing and lactation consultation professions are identifying and addressing the role of musculoskeletal issues such as positioning the infant at the breast[9,10] and soft tissue dysfunctions, like ankyloglossia (tongue-tie)[11] and congenital torticollis[9] as causes of SIB.[3] Providers across disciplines are beginning to emphasize correcting those factors that may interfere with infants' suckling[8] leading to decreased breastfeeding for optimal infant nutrition.

To further identify factors affecting SIB, clinicians may utilize outcome assessment tools (OATs) to identify effective treatments and practices. Identifying and understanding factors important to patients allows clinicians to assess the quality of services provided.[12] Outcome measures focus on identifying the effect of a condition on a patient's health status as well as the effectiveness of treatment strategies on that condition. The Musculoskeletal Infant Breastfeeding Assessment Questionnaire (MIBAQ) is an outcome measure

designed to address condition-specific detail related to SIB in infants.

Hawk et. al[8] presented the original MIBAQ form where 23 questions over four domains (i.e., latching, sucking, swallowing, and symmetry) and one global domain were identified as related to the mechanics of nursing. However, in the study Hawk et. al[8] recognized five questions where greater than 50% of participants marked "never or seldom." The investigators believed the items were either non-relevant or unable to be understood by the patients and chose to streamline the document. In response, 10 items were removed from the original MIBAQ form leading to the development of the MIBAQ-S form. The purpose of this multi-site cohort study was to present a streamlined version of the MIBAQ and assess outcomes of manual therapy interventions used to relieve symptoms of SIB.

Methods

This was an observational pilot cohort study conducted in multiple chiropractic clinics in the U.S. and Canada. Its purpose was to assess one-week outcomes of manual therapy by chiropractors for infants with musculoskeletal dysfunction and suboptimal infant breastfeeding (SIB), using the Musculoskeletal Infant Breastfeeding Questionnaire-Short Form (MIBAQ-SF).[8]

Informed consent

The lead institution's Institutional Review Board approved the project prior to data collection. Participating mothers signed an informed consent form prior to completing forms. All other data were collected anonymously using ID numbers only.

Participating clinics

We invited Doctors of Chiropractic (DCs) who self-reported experience in treating infants with suboptimal breastfeeding to participate. The resulting sample of convenience was composed of those DCs who agreed to participate.

Participating maternal-infant dyads

Participating clinics were instructed to enter maternal-infant dyads into the study using these eligibility criteria:

Inclusion criteria: Consecutively presenting mothers of currently breastfeeding infants age ≤ 3 months presenting for nursing difficulties at the participating clinic during the 2-month study period.

Exclusion criteria: Mother declining participation by refusal to fill out the forms.

Data Collection

All data besides consent-to-participate forms were collected electronically utilizing SurveyMonkey (non-HIPPA compliant software). Participants (eligible mothers of infant patients) completed medical history and pre-treatment MIBAQ-SF forms. Treating clinicians filled out a pre-treatment form providing information of previous and concurrent treatment of the infant and a post-treatment form providing information on treatment and discharge. The office staff in each participating office provided the SurveyMonkey (v.25) link to the patients at intake and at a one-week follow-up interval. Data were downloaded from SurveyMonkey by the lead institution electronically at the end of the study period.

Data collection period

The data collection period was three months. Data were collected immediately before the infant's first visit, prior to treatment, and at a one-week interval following the first visit while attending a subsequent visit. The determination to utilize a one-week interval was based on prior experience and the investigators' clinical experience that some improvement would likely be apparent at one week.

Outcome measures

We assessed outcomes using two instruments:

1. MIBAQ-SF (Musculoskeletal Infant Breastfeeding Assessment Questionnaire–Short Form). We based this on the original MIBAQ,8 deleting questions to which respondents had indicated "never" or that duplicated questions in the same domain, to streamline the form to facilitate mothers responding. This resulted in 13 questions covering the domains related to the mechanics of nursing (latching, sucking, swallowing, and symmetry) and one global domain (Table 1). The MIBAQ-SF, like the MIBAQ, uses a Likert scale of 0-3, where 0=never or seldom; 1= sometimes; 2=often; 3=very often or always.

Domain	Item
Latching	1. Slips off nipple
	2. Latches on the tip of nipple area only
	3. Bites or chomps on nipple
	4. Can't open mouth widely
Sucking	5. Starts and stops nursing during a feeding
	6. Falls asleep during feeding
	7. Sucking sounds not rhythmic
	8. Baby does not empty breast when feeding
Swallowing	9. Milk spills out of mouth, gags or makes clicking or whistling sound while nursing
	10. Excessive gas, burping, spitting up
Symmetry	11. Difficulty latching on one breast more than the other
Global	12. Turns head to one side more frequently or more easily
Global	13. Wants to nurse almost constantly

Table 1. MIBAQ-SF domains and items.

2. Patient Global Impression of Change (PGIC). We included this instrument to assess the MIBAQ-SF's sensitivity to clinical change. The PGIC's validity and reliability have been established for measuring clinical response to treatment, and it is widely used in healthcare research.[13,14] The PGIC is administered post-treatment only. However, because it is a global measure, it is designed to be combined with domain-specific instruments. It consists of a seven-item Likert scale about the patient's self-report of their response to treatment in which 7=very much better, 6=much better, 5=a little better, 4=no change, 3=a little worse, 2=much worse and 1=very much worse. We adapted the PGIC for this study by asking the mother to check the box for how her baby's breastfeeding is now, compared to before treatment at this office.

The pre- and post-forms were identical except that the Patient Global Impression of Change was included in the post-MIBAQ. Investigators took a conservative approach to scoring, imputing all missing values as 0, which would indicate that the symptom was never/seldom present.

Data management and analysis

Data were downloaded directly to SPSS (v.25) from SurveyMonkey for statistical analysis. Total MIBAQ scores were computed by summing all 13 items' responses; the possible range would be 0-39, with lower scores indicating fewer symptoms of nursing dysfunction. Total pre- and post-MIBAQ-SF scores were compared using a paired t-test. Further analysis included comparison of the overall change score (difference between mean pre- and post-MIBAQ total scores) to the mean PGIC score using a Pearson correlation.

Results

A total of 72 participants (mother-infant dyads) from six chiropractic offices completed the pre-survey. There were 34 (47%) boy and 27 (38%) girl infants, with 11 infants with missing data for gender (15%). For previous and concurrent care, 50 (69%) mothers reported seeing a lactation consultant (11 missing data, 15%); 15 of these were still seeing the consultant and 35 were not. Forty-one (57%) infants had previously been diagnosed with tongue-tie and 23 (32%) had received medical treatment for it. The six participating DCs for the 72 participating dyads completed post surveys on treatment and discharge for 54 (72%).

Total MIBAQ-SF score*			
Pre- 18.8	Post- 12.2	Pre- to post- change 6.6	Significance p= .000

*Maximum score= 39;
lower score indicates better nursing function.

Table 2. Pre-post MIBAQ-SF scores from paired samples t-test (n=35).

For outcome measures, 35 (49%) mothers completed both pre- and post-survey and six clinicians provided post-treatment information for 53 (74%) of patients. For participants, the MIBAQ scores improved highly significantly ($p < .000$) from pre- to post (Table 2). The MIBAQ-SF change scores were significantly correlated with the PGIC (Pearson correlation=.408; p=.021). Table 3 shows the PGIC scores; the mean score was 5.5 one week from baseline. Clinicians who provided data reported the average number of treatments within the 1-week timeframe for each patient was 2.4 treatments.

1. 18% – very much better
2. 30% – much better
3. 36% – a little better
4. 14% – no change
5. 2% – a little worse
6. 0% – much worse
7. 0% very much worse

Table 3. PGIC Scores at One Week from Baseline.

Discussion

Suboptimal infant breastfeeding is a multifactorial issue as varied as the patients themselves.[3] Musculoskeletal issues have been identified by authorities in the lactation counseling profession as interfering with breastfeeding.[3,15,16] Measuring latch is completed by a lactation consultant and there are five screening tools utilized by lactation specific professionals: BREAST Feed Observation Form, Lactation Assessment Tool, LATCH Scoring System, Mother-Baby Assessment Tool, and Mother-Infant Breastfeeding Progress Tool. Chiropractors do not have the ability to complete a thorough assessment of latch, indicating a need for professionals in the chiropractic realm to have an assessment tool. The initial MIBAQ[8] was introduced as a method of measuring SIB without measuring latch, creating a method for practitioners to gain inter- and intra-personal information regarding breastfeeding beyond lactation consultants.

Following data analysis of the original MIBAQ[8] investigators believed the form could be streamlined, leading to the development of the MIBAQ-SF. The MIBAQ-SF appears to be a feasible outcome assessment tool to use in a chiropractic clinic specializing in infant care for the purposes of treating SIB. The MIBAQ-SF also appears to be sensitive to change. In this pilot study we compared pre- and post-treatment scores to the PGIC a known reliable, sensitive, and previously validated tool.[13,14] The PGIC is a global measure of change, and recommendation for utilizing the measure is to use it in combination with a condition specific measure such as the MIBAQ-SF.[13,14]

This study was not to determine effective strategies for management of SIB, but to investigate the outcome measure, MIBAQ-SF, and determine if it is valid and sensitive to change. Further investigation into specific strategies utilizing the MIBAQ-SF is an option for a later date.

During this study 50% of mothers completed both the pre- and post- forms. Reasons for the low follow-up rate were not provided by practitioners, but it is likely patients were not presented with the correct link to the online survey at the one-week follow-up. Follow-up was also made difficult using a non-HIPPA compliant form of SurveyMonkey. The decision to use the two number month and date of birth for the infant caused difficulty through multiple patients with the same date of birth and in one case a pair of twins. Further study would necessitate the use of a HIPPA compliant format that would allow for data collection linked to each patient and doctor.

Strengths

Instrument appeared easy to use and demonstrated clinical responsiveness.

Limitations

Anonymous data collection was more difficult due to the use of a non-HIPPA compliant version of SurveyMonkey. Duplicate patient identification numbers were utilized by mothers at numerous clinics, and follow-up data analysis included linking patient identification numbers to clinician identification numbers. Future research would necessitate the use of a HIPAA compliant format for data collection.

Another limitation to the study was inconsistent data recording from clinicians as a low number of clinicians provided data during the two-month study period. Eighteen clinicians opted to participate in the study and seven provided data for 72 participating mothers. Two clinicians communicated they did not have patients fitting inclusion criteria during the study timeframe. There is no accounting for failure to provide data from the other nine clinicians. Future research might include a post-study questionnaire to determine factors that may have influenced clinician compliance.

Although not the main purpose of the study, clinicians were asked to provide data regarding the number of treatments each infant received from pre- to post- reporting of the MIBAQ-SF. Conclusions regarding the optimal number of treatments necessary to report a change in the MIBAQ-SF were unable to be drawn during this study due to insufficient reporting of the number of treatments during the study period. The average number of treatments provided to participants was 2.4. However, only six clinicians provided post-treatment information on 53 patients (74%). Future research should include the use of the MIBAQ-SF combined with the number of treatments provided to aid clinicians developing optimal goals for patients and their mothers.

Conclusions

These results demonstrate that the MIBAQ appears to be clinically responsive to changes in SIB-related symptoms and could facilitate larger practice-based research studies of infants with musculoskeletal dysfunction contributing to SIB. In this study, significant clinical change was reported by mothers of infants with SIB utilizing the MIBAQ-SF after one week of chiropractic manual therapy.

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References:

1. CDC Dietary Guidelines https://www.dietaryguidelines.gov/sites/default/files/2020-12/Dietary_Guidelines_for_Americans_2020-2025.pdf#page=65 (n.d.). Accessed May 13, 2022

2. World Health Organization. *Infant and young child feeding: Counselling cards for health workers*. World Health Organization. https://www.who.int/publications-detail-redirect/WHO-HEP-NFS-21.45 (n.d.). Accessed May 13, 2022,

3. Hawk C, Minkalis, A Webb, C Hogan O, & Vallone S. Manual Interventions for Musculoskeletal Factors in Infants with Suboptimal Breastfeeding: A Scoping Review. *Journal of Evidence-based Integrative Medicine*, 2018;23:1-12

4. Bartick MC, Schwarz EB, Green BD, Jegier BJ, Reinhold AG, Colaizy TT, Bogen DL, Schaefer AJ, & Stuebe AM. Suboptimal breastfeeding in the United States: Maternal and pediatric health outcomes and costs. *Maternal & child nutrition* 2017;13(1), e12366.

5. Zakarija-Grkovic I, Šegvic O, Vuckovic Vukušic A, Lozancic T, Bozinovic T, Cuze A, Burmaz T. Predictors of suboptimal breastfeeding: an opportunity for public health interventions, *European Journal of Public Health*, 2016;26(2): 282—289.

6. Genna CW. Breastfeeding infants with congenital torticollis. *J Hum Lact*. 2015; 31:216—220.

7. Increase the proportion of infants who are breastfed at 1 year - MICH 16 - Healthy People 2030. https://health.gov/healthypeople/objectives-and-data/browse-objectives/infants/increase-proportion-infants-who-are-breastfed-1-year-mich-16 (n.d.). Accessed May 13, 2022,

8. Hawk C, Vallone S, Young J, Lavigne V. Development of an outcome assessment instrument for suboptimal breastfeeding in infants with musculoskeletal dysfunction. *Journal of Clinical Chiropractic Pediatrics*.2020; 19(1):1621-1628.

9. Genna CW, editor, ed. Supporting Sucking Skills in Breastfeeding Infants., 3rd ed Burlington MA: Jones & Bartlett; 2017.

10. Marmet C, Shell E. Therapeutic positioning for breastfeeding in: Genna CW, editor., ed. Supporting Sucking Skills in Breastfeeding Infants. Burlington, MA: Jones & Bartlett; 2017:399—416.

11. Coryllos E, Genna CW, Fram JL. Minimally invasive treatment for posterior tongue-tie (the hidden tongue-tie) In: Genna CW, editor., ed. Supporting Sucking Skills in Breastfeeding Infants. Burlington, MA: Jones & Bartlett; 2017:269—278.

12. Snyder AR, Parsons JT, Valovich McLeod TC, Curtis Bay R, Michener LA, & Sauers EL. Using disablement models and clinical outcomes assessment to enable evidence-based athletic training practice, part I: disablement models. *Journal of Athletic Training*, 2008;43(4), 428—436.

13. Rampakakis E, Ste-Marie PA, Sampalis JS, Karellis A, Shir Y, Fitzcharles MA. Real-life assessment of the validity of patient global impression of change in fibromyalgia. *RMD Open*. 2015;1(1): e000146.

14. Scott W, McCracken LM. Patients' impression of change following treatment for chronic pain: global, specific, a single dimension, or many? *J Pain*. 2015;16(6):518-526.

15. Coryllos E, Genna CW, Fram JL. Minimally invasive treatment for posterior tongue-tie. In: Genna CW, ed. *Supporting Sucking Skills in Breastfeeding Infants*. Burlington MA: Jones and Bartlett; 2017:269-278.

16. Ghaheri BA, Cole M, Fausel SC, Chuop M, Mace JC. Breastfeeding improvement following tongue-tie and lip-tie release: A prospective cohort study. *Laryngoscope*. 2017;127(5):1217-1223.

Sleep in Autistic Children and Impact on Parental Stress

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ABSTRACT

The aim of this study was to examine the impact sleep quality of autistic children had on parental stress levels. Parental report was collected from five mothers who had children who had previously been diagnosed on the autism spectrum. Parents were recruited from a small sensory gym while their children were participating in class. The modified Children's Sleep Habits Questionnaire (CSHQ) and Parental Stress Scale were utilized to collect data. Consistent with previous research it appeared that sleep problems in autistic children correlated with increased parent stress levels as reported on the Parental Stress Scale. Maladaptive bedtime behaviors had a significant correlation with parent's stress levels compared to other aspects of the modified CSHQ. The results of this study should be utilized with previous research to provide evidence for the inclusion of sleep as an intervention focus for autistic children. Interventions aimed at helping these children improve their quality of sleep could help decrease the amount of stress seen in parents.

Key terms: autism, autism spectrum disorder, ASD, sleep, parent, parent stress.

Introduction

Sleep in Autistic Children and Impact on Parental Stress

Sleep is a necessity for all human beings and is required to adequately function in daily life. Sleep quality can impact one's physical health, quality of life, safety, and mental health.[1] Without adequate sleep, people may have difficulty controlling their emotions and behavior during the day. For children, this may lead to difficulties in school.

Autism Spectrum Disorder (ASD) is a prevalent disorder impacting 1 in 44 children in the United States.[2] ASD is characterized as a developmental disability caused by differences in the brain that lead to communication, social, and stereotyped behavioral problems impacting the individual's daily life.[3] The disorder is described as a spectrum disorder as children can benefit from tiered support in a variety of skill areas. ASD is usually diagnosed in children around age two but can also occur earlier or later in development depending on symptom severity.[3] The autism community has started to recognize use of identity-first language (autistic child) to be used instead of person-first language to fully embrace autistic identity. An autistic child can display problems in different settings such as school versus at home. While ASD can significantly impact a child's life, it can also have a unique impact on the lives of the child's parents.

Considering the amount of care autistic children require and the energy parents may expend communicating with their child, Dabrowska and Pisula (2010) found parents of autistic children appear to have a higher level of stress in comparison to parents of children with Down Syndrome (DS) or typical development (TD).[4] Differences between parents of autistic children and DS included stressors related to dependency ($p>.0001$), life span care ($p<.0001$), and limits on family opportunity ($p<.0001$).[4] Having an autistic child appears to lead to additional stress surrounding their care and reliance on a parental figure. Different characteristics and the severity of the child's manifestation of ASD can also impact parental stress. Communication and social skills appear to be significant indicators for higher parental stress levels because of the impact it has on the child-parent bond.[5] With the increased prevalence of ASD, it will be important to address the connection of stress and sleep in both the autistic child and the parent.

Problem Statement

Poor sleep patterns experienced in autistic children, have been linked to increased behavioral problems during the day.[6] Autistic children who had poor sleep quality showed an increase in hyperactivity, physical aggression, inattention, and irritability.[6] This escalation in symptoms exacerbated the parent's stress level leading to more difficulties for both the child and the parent. The relationship between poor sleep of the child, increased problem behaviors, and parent's stress becomes a perpetual cycle. The prolonged stress can create health problems for the parents.

Autistic children often exhibit sleep difficulties which increase maladaptive daytime behaviors and this impacts parental stress leading to poor health outcomes for parents. Increased, prolonged parental stress can be related to decreased health-related quality of life (QoL).[7] In comparison to parents of children with TD, health related QoL for parents of autistic children was diminished, which may have been impacted by the increased daytime behaviors seen in autistic children who had poor sleep quality. There appears to be a high correlation of stress with the increased number of health-related problems for parents with autistic children. According to Reed et al. (2016), parents of autistic

children also reported a higher amount of physical health problems related to immune function.[7] High stress can also lead to cardiovascular disease which perpetuates the strain on these parents.[8]

Methodology

This literature review includes scholarly, peer-reviewed articles found from multiple databases. Key words during searches included autism, autism spectrum disorder, parent stress, sleep, family stress, sleep quality, and sensory processing. Sources were included that included scientific information and focused on autistic children and their parents' stress levels. Sources were excluded that focused on autistic adults' sleep quality, sibling stress, or personal reports. Data were summarized in narrative to explore the impact sleep quality in autistic children has on parents' stress levels.

Literature Review

Sleep in Autistic Children and Typical Development

Many studies have looked to examine if quality of life is impacted in a variety of ways in autistic children. The sleep quality of these children is an emerging area of interest. Researchers have begun to investigate the differences in sleep quality and behavior in autistic children and those with typical development (TD). Sounders et al. (2009) conducted a descriptive cross-sectional study to examine the prevalence of sleep problems in autistic children and TD.[9] Parent report measures and actigraphy, an electronic device measuring objective sleep data, were utilized to gather data. Autistic children showed significant differences on sleep behaviors such as sleep terrors ($p=.009$), bed wetting ($p=.035$), and bruxism ($p=.002$).[9] The study revealed children in the ASD group had a 66.1% prevalence rate for sleep disturbances, while the children with TD only had a 45% rate.[9]

In a study conducted by Krakowiak, Goodlin-Jones, Hertz-Picciotoo, and Croen (2008), similar differences between autistic children and TD were also found.[10] Sleep problems were seen "frequently" ($p<.0001$) or "always" ($p<.05$) in 53% of autistic children compared to 32% of children with TD. Autistic children appeared to sleep on average an hour less than their peers with TD ($p<.0001$). The study also found more autistic children (24%) had problems with onset of sleep compared to children with TD (9%). Night wakings were also more common for the autistic children than the children with TD ($p=.001$).[10] Both studies revealed the significant difference in sleep behavior and quality in children with and without ASD.

Characteristics of Sleep in Autistic Children

Researchers have begun to examine what leads to autistic children having poor sleep quality and what areas of sleep are impacted. The Children's Sleep Habits Questionnaire (CSHQ) is a parent report measure examining nine domains of sleep behavior such as bedtime resistance and sleep onset delay.[11] With many researchers utilizing the CSHQ, similarities between studies have emerged.[11-13] Characteristics of sleep that are problematic for many autistic children are described below.

Sleep Characteristics

When (what time) a child falls asleep and how long it takes them to fall asleep is often included in screenings for sleep problems. Parents of autistic children list sleep onset as one of their major concerns when it comes to their children's sleep habits.[11] Autistic children often have problems falling asleep often noted on parental reports on the CSHQ.[11] Bedtime resistance is also often observed in autistic children. Malow et al. (2006) additionally found bedtime resistance as a major problem for autistic children who were classified as poor sleepers ($p<.0421$).[11] In another study, Liu et al. (2006) reported 56.3% of the autistic children in their study displayed bedtime resistance.[12] These children were also more likely to have problems with insomnia and morning rise. Characteristics associated with bedtime resistance included allergies, hypersensitivity, bedsharing, and father's sleep problems.[12]

Insomnias (56.3%) and parasomnias (53.3%) were also extremely prevalent in autistic children.[12] Conditions such as asthma were associated with elevated scores for insomnias, while younger age, use of medication, and bedtime ritual were linked with frequent parasomnias. Gastrointestinal problems were associated with higher prevalence of insomnias and parasomnias.[12] Factors which did not appear to impact the child's sleep problems included type of ASD, severity of diagnosis, gender, bedtime and rise time, number of family members, environmental factors, married status of parents, and maternal education.[12] Understanding the common sleep characteristics seen in autistic children can help researchers understand what may be causing the increase in sleep problems.

Sensory processing

Autistic children tend to have problems with sensory processing which can lead to increased sleep problems.[13,14] Both studies have examined the impact a sensory processing problem has on the child's sleep quality. Reynolds et al. (2012) reported 81% of children in their study had a significant score in at least one quadrant on the Sensory Profile.[14] When the child scored in the sensation avoiding quadrant, there was significant correlation with sleep problems ($p=.011$). These children may find it difficult to filter sensory information which can lead to trouble calming down to go to bed and to sensory over-responsiveness.[14] Mazurek and Petroski (2015) also examined sensory processing and sleep in autistic children.[13] They found significant correlation between sensory over-responsiveness and increased scores on the CSHQ ($p<.001$). This arousal dysregulation may

contribute to the sleep problems witnessed in autistic children. Sensory over-responsiveness may interfere with sleep quality due to the sensory stimuli present during the sleep environment such as noise, light, and temperature.[13] Sensory processing is often seen in autistic children and further research on its connection to sleep problems is needed.

Anxiety

Anxiety levels in autistic children are also associated with increased maladaptive sleep behaviors. A study was conducted to examine the impact of anxiety on sleep problems. According to Mazurek and Petroski (2015), anxiety appeared to have a significant negative impact on bedtime resistance ($p<.001$), sleep-onset delay ($p<.001$), sleep duration ($p<.001$), sleep anxiety ($p<.02$), and night wakings ($p<.001$).[13] Anxiety seemed to impact many of the common sleep problems experienced by autistic children. Malow et al. (2006) also examined anxiety levels in autistic children.[11] The Child Behavior Checklist (CBCL) was used to examine anxious symptoms expressed by the children per parental report. They found there was a significant increase in anxious symptoms in autistic children who had poor sleep problems ($p<.0156$)[11]. Anxiety seems to frequently occur in autistic children and appears to relate to decreased sleep quality.

Impact of Sleep Quality on Autistic Children

Daytime behaviors. When a child has decreased sleep quality, they are more likely to display increases in maladaptive daytime behaviors. For autistic children, these increased behaviors may be displayed as frequent and more severe characteristics of ASD. According to Tudor, Hoffman, and Sweeney (2012), "Sleep disturbances, problems with sleep onset, and short sleep duration were associated with all autism symptoms as well as overall autism severity" (p. 258).[15] Children who reported decreased quality of sleep appeared to display more stereotyped behaviors, social impairments, and communication difficulties. The decreased quality of sleep appeared to increase the severity of the characteristics of ASD.[15]

Mazurek and Sohl (2016) also examined how decreased sleep impacted how the child behaved during the day.[6] In their study, the behavioral characteristics of aggression, hostility, inattention, and hyperactivity were examined in 81 autistic children. When investigating these behaviors in relation to scores on the CSHQ, autistic children appeared to show increased maladaptive behaviors. Parents who reported their children had increased daytime sleepiness, also noted increased hyperactivity ($p<.01$). Nighttime awakenings were significantly correlated with increased physical aggression ($p<.0001$), inattention ($p<.01$), and hyperactivity ($p<.05$). Children who had high levels of sleep anxiety appeared to be more irritable during the day. The study also noted sleep duration and parasomnias negatively impacted all four behaviors.[6] Children who display sleep quality impairments appear to have increased ASD symptomology severity and increased maladaptive behaviors during the day.[6,15]

Quality of life. Sleep quality can also negatively impact quality of life (QoL). Delahaye et al. (2014) examined the impact of sleep problems in autistic children on their health-related quality of life (HRQoL).[16] Results from the study indicated lower HRQoL in all domains for autistic children compared to the normative population. Physical functioning, psychosocial function, and total functioning were significantly lower for autistic children ($p<.0001$). The children's total score on the CSHQ correlated with decreased total and psychosocial scores of HRQoL ($p<.0001$). The child's sleep duration appeared to have a significant correlation on the total and psychosocial scores of HRQoL ($p<.001$). This decrease in HRQoL present in these autistic children can have an impact on their daily lives. Delahaye et al. (2014) make note of this connection to emphasize the role poor sleep quality can have on the QoL of autistic children.[16]

Impact of Child's Sleep on Parents

Sleep. When an autistic child sleeps poorly, it appears to impact the parent's sleep quality as well.[17-19] Meltzer (2008) conducted a study examining the difference in sleep of parents with autistic children and TD.[19] Participants included 20 families with autistic children and 15 families of children with TD. Metlzer (2008) examined the sleep of the parents by utilizing the Pittsburg Sleep Quality Index (PSQI), actigraphy, and sleep diaries.[19] The study revealed parents of autistic children had poorer wake time ($p<.01$) and actual sleep time ($p<.05$) compared to parents with TD children. Fathers of autistic children reported earlier wake times ($p<.01$) and shorter sleep duration ($p<.01$) compared to mothers from the same family.[19] This may be due to factors other than the child's diagnosis, such as work schedules. Mothers reported significant differences on the scales of actual sleep time ($p<.001$), sleep efficiency ($p<.05$), and longest sleep period ($p<.05$) when compared to mothers of TD children.[19] It is to be expected if a autistic child does not have quality sleep behaviors, it would impact the parent's sleep as well. Limitations from Meltzer's study included lack of information regarding the child's sleep habits to examine if a relationship existed between the child and parent's sleep quality.[19] Hodge et al. (2013) examined the relationship between child and parent sleep quality and found the quality of a child's sleep is a significant indicator for impaired maternal sleep ($p<.001$).[18] When a parent has poor sleep habits, they can be at an increased risk for higher stress levels.

Stress level. Parents of autistic children are already at an increased risk of higher stress levels.[4,5] Since the autistic child has difficulties with sleep quality, the purpose of this

study is to examine if the child's poor sleep quality impacts the increased stress seen in their parents. Levin and Scher (2016) examined the relationship between child's sleep quality and parent's stress level in families with children with and without ASD.[20] The autistic children displayed more frequent sleep problems compared to children with TD (p=.02). The sleep domains of bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, and parasomnias were significantly correlated with increased maternal stress levels.[17,20] Mothers expressed increased worry about their child's safety during the night compared to mothers of children with TD.[20]

Hodge et al. (2013) also examined the direct impact of the sleep quality in an autistic child on maternal mental health.[18] Their study examined 180 mothers and their child with half of the participants having autistic children. In this study, the quality of a child's sleep was a significant indicator for maternal mental health and maternal stress (p<.001). When examining the participants who had children with TD, there also appeared to be a correlation between the quality of sleep of the TD child and mother leading to higher levels of maternal stress (p<.001). Mothers of autistic children in the study also reported they viewed poor sleep quality as minor compared to other aspects of their child's disability as it contributes to their stress level.[18] While it appeared there was a relationship between the child's sleep quality and maternal stress levels, it did not seem sleep problems were major stressors for mothers of autistic children.

Summary

An autistic child appears to have poor sleep habits when compared with children with TD.[9,10] This decreased sleep quality seems to impact the child's behaviors during the day and overall quality of life.[6,15] The poor sleep appears to also impact more than just the child, but also impacts parents. Parents of autistic children are already displaying higher stress levels than parents of children with TD.[4,5] When their child has poor sleep, it appears to impact parents by increasing their stress levels, decreasing their mental health, and impairing their sleep quality.[17-19] This study aims to explore the relationship between the sleep quality of an autistic child and the impact it has on parental stress levels to examine if sleep is a major contributor to parental stress.

Method

Participants

Five families participated in the study. All participants who filled out the questionnaire packet consisted of married mothers of autistic children. Mother's age ranged from 38 to 43 (M=40.2). Three of the mothers reported full-time or part-time employment, while two mothers were considered stay-at-home. The children's age ranged from 4.5 to 12 (M=8.3). All the children were participants in the sensory program and were male.

Questionnaires

The modified Children's Sleep Habits Questionnaire (CSHQ) was created to distinguish sleep differences in autistic children.[21] It is a parent report measure examining a child's bedtime, sleep behavior, wakings during the night, and morning wake up. The measure consists of 22 items to be ranked on a scale from 0 (never) to 7 (always). The authors found parts of the Children's Sleep Habits Questionnaire to be irrelevant when used with the ASD population. The Modified CSHQ requires additional testing to examine test-retest reliability, validity, and relevance, and to examine its impact on typically developing children. The original CSHQ developed by Owens, Spirito, and McGuinn (2000) has been utilized in many studies to examine sleep in children.[22] Psychometric properties of the original CSHQ include .78 internal consistency rate when examining a clinical sample. The measure also had good validity which was confirmed with the clinical sample having significantly higher or worse scores on all subscales compared to the community group (p<.001). The original CSHQ also showed stable test-retest reliability.[22]

The Parental Stress Scale is a self-report measure examining 18 items about varying aspects of parenthood both positive and negative.[23] Participants rate each item on a five-point scale from 1 (strongly disagree) to 5 (strongly agree). A higher score on the scale correlates with a higher level of stress. The scale has been examined for both parents of children with typical and atypical development. The scale has internal reliability of r=.83 and test-retest reliability of .81.[23] In a study conducted by Zelman and Ferro (2018), the scale was examined for use with families of children with chronic health conditions.[24] Internal consistency reliability was reported as a=.84, and the measure reported good validity in comparison with another standardized assessment.[24]

Procedure

This study received Institutional Review Board (IRB) approval from Bay Path University. Approval from a local community center was sought out as they were running a sensory group for children with and without ASD. The program met daily after school to provide sensory experiences to the children involved. Parents sit outside of the sensory gym and were asked if they would be willing to fill out a few surveys. Parents who consented to the study were given the demographic form, modified CSHQ, and the Parental Stress Scale. Parents returned these forms when they were done to the investigator. Scoring of the Parental Stress Scale and CSHQ were completed by the researcher, and SPSS was used to run a two-tailed Pearson correlation test.

Results

After parent completion of the questionnaire packets, analysis was completed. All the children had their own bedrooms except for one child who shared his room with a twin. Time spent on bedtime routines with the child varied greatly among the participants. Parents reported spending as little as 20 minutes to over an hour with their child to get them to fall asleep. Most participants had a set bedtime routine which may have included putting on pajamas, brushing teeth, reading a story, watching a television show, and showering/bathing. Only one parent reported their child took naps rarely, while the other children never engaged in napping. Three parents reported being concerned about their child's bedtime routine or sleep quality. The modified CSHQ scores can range from 0 to 88. For this study, the average score on the modified CSHQ ranged from 17 to 32 (M=23.6). Child's age was not significantly correlated with the child's scores on the modified CSHQ. Child sleep duration also did not appear to impact the parent's reported score on the CSHQ. The bedtime subscale had the highest scores on the modified CSHQ when compared to the subscales of sleep behavior, waking, and morning wake.

When parents were asked about their sleep patterns, three parents reported having a difficult time falling asleep or staying asleep. Scores on the Parental Stress Scale can range from 18-90, with higher scores indicating greater stress. In this study, parent's scores ranged from 41 to 55 (M=47.6). Parents agreed with the statement, "The major source of stress in my life is my child(ren)" in four of the five participants. Parents also reported they agreed or strongly agreed (80%) having children has left them with little time and flexibility in their life. The majority of the parents (80%) agreed or strongly agreed they are satisfied as a parent and find their children enjoyable. Parent's scores on the Parental Stress Score were significantly correlated with increased scores on the bedtime subscale of the modified CSHQ ($p<.059$). Parental Stress Scores were not associated with parent age, child age, parent concern, parent sleep, child's total score on the modified CSHQ, or child's sleep duration.

Discussion

From the previous literature, autistic children often have increased sleep difficulties.[9,10] From the research study, children did show increased difficulties with the bedtime behavior subscale on the modified CSHQ. While the sample size was small, there was evidence to support the presence of sleep difficulties in autistic children. The current study failed to confirm the sleep differences between children with ASD or TD since a control group was not utilized. Children also did not appear to have decreased sleep duration unlike findings from previously stated research. Since the current study was not focused on children's sleep on their daytime behavior, there were no significant conclusions sleep has an impact on their behavior, but much research has shown a correlation between poor sleep quality in autistic children and an increase in maladaptive daytime behavior.

The current study aimed to examine the relationship between sleep quality in autistic children and the impact it has on parental stress levels. While the participants did not report significant sleep differences in the children, the participants did express a slight increase in parental stress. The participant's score on the Parental Stress Scale was significantly correlated with the child's score on the bedtime behavior subscale. This confirms findings in which sleep quality in autistic children does have an impact on parental stress levels.[17-19] Similar to the findings of Hodge et al. (2013), parents did not appear to rank their child's sleep difficulties as a significant contributor to their stress level, but the majority of participants in this study did appear to find their child's sleep quality concerning.

Limitations

There were limitations in this study. The sample size (n=5) was very small and was considered a convenience sample based on which parents brought their children to the sensory program. The participants also consisted of only mothers, and the data cannot be generalized to fathers of autistic children. With such a small sample size, the results can only attempt to support the evidence already published regarding child's sleep and parent stress. The data were also collected by parent report and consisted only of subjective data. Future research would have to include objective data to examine the child's sleep quality and more in depth questionnaires regarding parental stress.

Conclusion

When working with an autistic child, it is also important to include the parent because they often have an increased level of stress. Since sleep is essential and considered an activity of daily living by the American Occupational Therapy Association, it is important for occupational therapy practitioners to address sleep in both the autistic child and their parent. Starting to examine interventions which could help an autistic child receive a better quality sleep may help reduce parental stress. Researchers have begun to examine the positive impact behavioral interventions have on sleep in autistic children. With interventions being developed to improve sleep quality in autistic children, reductions in parent's stress levels may appear decreasing the negative impact stress has on parent's quality of life.

References:

1. Touchette E, Petit D, Seguin JR, Boivin M, Tremblay RE, Montplaisir J . Associations between sleep duration patterns and behavioral/cognitive functioning at school entry. *Sleep*. 2007; 30(9): 1213-1219. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/articles/PMC1978413/pdf/aasm.30.9.1213.pdf.

2. Maenner MJ, Shaw KA, Bakian AV, et al. (2021). Prevalence and characteristics of autism spectrum disorder among children aged 8 years – *Autism and developmental disabilities monitoring network*. 2021; 70(No. SS-11): 1—16. Retrieved https://www.cdc.gov/mmwr/volumes/70/ss/ss7011a1.htm?s_cid=ss7011a1_w.

3. American Psychiatric Association (APA). Neurodevelopmental disorders. *Diagnostic and Statistical Manual of Mental Disorders*, Fifth Edition. Washington, DC: APA Publishing; 2013. Retrieved from https://doi.org/10.1176/appi.books.9780890425596.dsm01.

4. Dabrowska A, Pisula E. Parenting stress and coping styles in mothers and fathers of pre-school children with autism and down syndrome. *Journal of Intellectual Disability Research*. 2010; 54: 266-280. doi:10.111/j.1365-2788.2010.01258.x.

5. Davis NO, Carter AS. Parenting stress in mothers and fathers of toddlers with autism spectrum disorders: Associations with child characteristics. *Journal of Autism Developmental Disorder*. 2008; 38: 1278-1291. doi: 10.1007/s10803-007-0512-z.

6. Mazurek MO, Sohl K. Sleep and behavioral problems in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders*. 2016; 46: 1906-1915. doi:10.1007/s10803-016-2723-7.

7. Reed P, Sejunaite K, Osborne LA. Relationship between self-reported health and stress in mothers of children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*. 2016; 46:934-941. doi:10.1007/s10803-015-2638-8.

8. Steptoe A, Kivimaki M. Stress and cardiovascular disease. *National Reviews Cardiology*. 2012; 9: 360-370. doi:10.1038/nrcardio.2012.45.

9. Souders MC, Mason TBA, Valladares O, Bucan M, Levy SE, Mandell DS, Weaver TE, Pinto-Martin J. Sleep behaviors and sleep quality in children with autism spectrum disorder. *Sleep*. 2009; 32(12): 1566-1578. Retrieved from https://academic.oup.com/sleep.

10. Krakowiak P, Goodlin-Jones B, Hertz-Picciotoo I, Croen LA, Hansen RL. Sleep problems in children with autism spectrum disorders, developmental delays, and typical development: A population-based study. *Journal of Sleep Research*. 2008; 17: 197-206. doi:10.111/j.1365-2869.2008.00650.x.

11. Malow BA, Marzec ML, McGrew SG, Wang L, Henderson LM, Stone WL. Characterizing sleep in children with autism spectrum disorders: A multidimensional approach. *Sleep*. 2006; 29(12), 1563-1571. Retrieved from https://academic.oup.com/sleep/article/29/12/1563/2709258.

12. Liu X, Hubbard JA, Fabes R A, Adam JB. Sleep disturbances and correlates of children with autism spectrum disorder. *Child Psychiatry and Human Development*. 2006; 37: 179-191. Retrieved from https://link.springer.com/journal/10578.

13. Mazurek MO.Petroski GF. Sleep problems in children with autism spectrum disorder: Examining the contributions of sensory over-responsivity and anxiety. *Sleep Medicine*. 2015;16: 270-279. Retrieved from http://dx.doi.org/10.1016/j.sleep.2014.11.006.

14. Reynolds S, Lane SJ, Thacker L. Sensory processing, physiological stress, and sleep behaviors in children with and without autism spectrum disorder. *OTJR: Occupation, Participation, and Health*. 2012; 32(1): 246-257. Retrieved from http://dx.doi.org/10.3928/15394492-20110513-02.

15. Tudor ME, Hoffman CD, Sweeney DP. Children with autism: Sleep problems and symptom severity. *Focus on Autism and Other Developmental Disabilities*. 2012; 27(4): 254-262. doi:10.1177/1088357612457989.

16. Delahaye J, Kovacs E, Sikora D, Hall TA, Orlich F, Clemons TE, Van Der Weerd, E, Glick L, Kuhlthau K. The relationship between health-related quality of life and sleep problems in children with autism spectrum disorder. *Research in Autism Spectrum Disorders*. 2014; 8: 292-303. Retrieved from http://dx.doi.org/10.1016/j.rasd.2013.12.015.

17. Hoffman CD, Sweeney DP, Lopez-Wagner MC, Hodge D, Nam CY, & Botts BH. Children with autism sleep problems and mothers' stress. *Focus on Autism and Other Developmental Disabilities*.2008; 28(3): 155-165. doi:10.1177/1088357608316271.

18. Hodge D, Hoffman CD, Sweeney DP, Riggs ML. Relationship between children's sleep and mental health in mothers of children with and without autism. *Journal of Autism and Developmental Disorder*. 2013; 43: 956-963. doi:10.1007/s10803-012-1639-0.

19. Meltzer LJ. Brief report: Sleep in parents of children with autism spectrum disorders. *Journal of Pediatric Psychology*. 2008; 33(4): 380-386. doi:10.1007/s11920-017-0782-x.

20. Levin A. & Scher A. Sleep problems in young children with autism spectrum disorders: A study of parenting stress, mothers' sleep-related cognitions, and bedtime behaviors. *CNS Neuroscience & Therapeutics*. 2016; 22(11): 921-927. doi:10.1111/cns.12651.

21. Katz T, Shui AM, Johnson CR, Richdale AL, Reynolds AM, Scahill L, Malow BA. Modification of the children's sleep habits questionnaire for children with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 2018; 48: 2629—2641. Retrieved from https://doi.org/10.1007/s10803-018-3520-2.

22. Owens JA, Spirito A, McGuinn M. The children's sleep habits questionnaire (CSHQ): Psychometric properties of a survey instrument for school-aged children. *Sleep*. 2000; 23(8): 1-9. Retrieved from https://academic.oup.com/sleep/article-pdf/23/8/1/21634304/230806.pdf.

23. Berry JO, Jones WH. The parental stress scale: Initial psychometric evidence. *Journal of Social and Personal Relationships*. 1995; 12: 463-472.

24. Zelman JJ, Ferro MA. The parental stress scale: Psychometric properties in families of children with chronic health conditions. *Family Relations: Interdisciplinary Journal of Applied Family Science* 2018; 67: 240-252. doi:10.1111/fare.12306.

25. Malow BA, Adkins KW, Reynolds A, Weiss SK, Loh A, Fawkes D, Katz T, Goldman SE, Madduri N, Hundley R, Clemons T. Parent-based sleep education for children with autism spectrum disorders. *J Autism Dev Disord*. 2014 Jan;44(1):216-28. doi: 10.1007/s10803-013-1866-z. PMID: 23754339; PMCID: PMC3818449.

Appendix B
Questionnaire Packet

Demographic Form

Thank you for participating in this study. Please take some time to fill out this demographic form along with the two attached questionnaires.

1. Age of person filling out this form: _____ 2. Relationship to child: _____

3. Marital status: _____

4. Employment status: Full-Time Part-Time Self-Employed Stay-at-Home Unemployed

5. Number of people living in household: _____ 6. Children in household: _____

7. Age of child involved in Sensiplay: _____ 8. Gender of child: _____

9. Child's grade in school: _____

10. Does the child have any medical diagnoses? _____

11. Does the child have his/her own bedroom? Yes No

12. How long does a bedtime routine take daily? _____

13. How often does the child sleep in his/her own bed? _____

14. What does the child's bedtime routine consist of? _____

---

15. Are you concerned by your child's bedtime routine or sleep quality? Yes No

16. On average, when do you fall asleep each night? _____

17. When do you begin your bedtime routine? _____

18. Do you have a difficult time falling asleep or staying asleep? Yes No

NICHD SECCYD—Wisconsin

CHILDREN'S SLEEP HABITS QUESTIONNAIRE (ABBREVIATED)

The following statements are about your child's sleep habits and possible difficulties with sleep. Think about the past week in your life when you answer the questions. If last week was unusual for a specific reason, choose the most recent typical week. Unless noted, check Always if something occurs every night, Usually if it occurs 5 or 6 times a week, Sometimes if it occurs 2 to 4 times a week, Rarely if it occurs once a week, and Never if it occurs less than once a week.

BEDTIME

Write in your child's usual bedtime: Weeknights _____:_____ am/pm

Weekends _____:_____ am/pm

	7 Always	5-6 Usually	2-4 Sometimes	1 Rarely	0 Never
1. Child goes to bed at the same time at night.	( )	( )	( )	( )	( )
2. Child falls asleep within 20 minutes after going to bed.	( )	( )	( )	( )	( )
3. Child falls asleep alone in own bed.	( )	( )	( )	( )	( )
4. Child falls asleep in parent's or sibling's bed.	( )	( )	( )	( )	( )
5. Child falls asleep with rocking or rhythmic movements.	( )	( )	( )	( )	( )
6. Child needs special object to fall asleep (doll, special blanket, stuffed animal, etc.).	( )	( )	( )	( )	( )
7. Child needs parent in the room to fall asleep.	( )	( )	( )	( )	( )
8. Child resists going to bed at bedtime.	( )	( )	( )	( )	( )
9. Child is afraid of sleeping in the dark.	( )	( )	( )	( )	( )

SLEEP BEHAVIOR

Write in your child's usual amount of sleep each day (combining nighttime sleep and naps): _____ hours and _____ minutes

	7 Always	5-6 Usually	2-4 Sometimes	1 Rarely	0 Never
10. Child sleeps about the same amount each day.	( )	( )	( )	( )	( )
11. Child is restless and moves a lot during sleep.	( )	( )	( )	( )	( )

NICHD SECCYD—Wisconsin

	7 Always	5-6 Usually	2-4 Sometimes	1 Rarely	0 Never
12. Child moves to someone else's bed during the night (parent, sibling, etc.).	( )	( )	( )	( )	( )
13. Child grinds teeth during sleep (your dentist may have told you this).	( )	( )	( )	( )	( )
14. Child snores loudly.	( )	( )	( )	( )	( )
15. Child awakens during the night and is sweating, screaming, and inconsolable.	( )	( )	( )	( )	( )
16. Child naps during the day.	( )	( )	( )	( )	( )
Write in the number of minutes the nap usually lasts: _____ minutes					

WAKING DURING THE NIGHT

	7 Always	5-6 Usually	2-4 Sometimes	1 Rarely	0 Never
17. Child wakes up once during the night.	( )	( )	( )	( )	( )
18. Child wakes up more than once during the night.	( )	( )	( )	( )	( )

MORNING WAKE UP

Write in the time child usually wakes up in the morning: Weekdays _____:_____ am/pm

Weekends _____:_____ am/pm

	7 Always	5-6 Usually	2-4 Sometimes	1 Rarely	0 Never
19. Child wakes up by him/herself.	( )	( )	( )	( )	( )
20. Child wakes up very early in the morning (or, earlier than necessary or desired).	( )	( )	( )	( )	( )
21. Child seems tired during the daytime.	( )	( )	( )	( )	( )
22. Child falls asleep while involved in activities.	( )	( )	( )	( )	( )

Parental Stress Scale

The following statements describe feelings and perceptions about the experience of being a parent. Think of each of the items in terms of how your relationship with your child or children typically is. Please indicate the degree to which you agree or disagree with the following items by placing the appropriate number in the space provided.

1 = Strongly disagree 2 = Disagree 3 = Undecided 4 = Agree 5 = Strongly agree

____ 1. I am happy in my role as a parent.

____ 2. There is little or nothing I wouldn't do for my child(ren) if it was necessary.

____ 3. Caring for my child(ren) sometimes takes more time and energy than I have to give.

____ 4. I sometimes worry whether I am doing enough for my child(ren).

____ 5. I feel close to my child(ren).

____ 6. I enjoy spending time with my child(ren).

____ 7. My child(ren) is an important source of affection for me.

____ 8. Having child(ren) gives me a more certain and optimistic view for the future.

____ 9. The major source of stress in my life is my child(ren).

____ 10. Having child(ren) leaves little time and flexibility in my life.

____ 11. Having child(ren) has been a financial burden.

____ 12. It is difficult to balance different responsibilities because of my child(ren).

____ 13. The behavior of my child(ren) is often embarrassing or stressful to me.

____ 14. If I had it to do over again, I might decide not to have child(ren).

____ 15. I feel overwhelmed by the responsibility of being a parent.

____ 16. Having child(ren) has meant having too few choices and too little control over my life.

____ 17. I am satisfied as a parent.

____ 18. I find my child(ren) enjoyable.

Berry, J. O., & Jones, W. H. (1995). The Parental Stress Scale: Initial psychometric evidence. Journal of Social and Personal Relationships, 12, 463-472.

Appendix C
Data Analysis

Correlations

Notes

Output Created		26-APR-2018 12:27:07
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Missing Value Handling	Definition of Missing	User-defined missing values are treated as missing.
	Cases Used	Statistics for each pair of variables are based on all the cases with valid data for that pair.
Syntax		CORRELATIONS /VARIABLES=Parentstressscale CSHQ Bedtime Sleepbehavior waking morningwake Parentage Childage Parentconcern Parentsleep childsleepduration /PRINT=TWOTAIL NOSIG /MISSING=PAIRWISE.
Resources	Processor Time	00:00:00.00
	Elapsed Time	00:00:00.00

Correlations

		Parentstressscale	CSHQ	Bedtime	Sleepbehavior	waking	morningwake	Parentage	Childage	Parentconcern	Parentsleep	childsleepduration
Parentstressscale	Pearson Correlation	1	.717	.863	-.228	-.133	-.272	-.735	-.019	-.017	-.254	-.286
	Sig. (2-tailed)		.173	.059	.712	.831	.657	.157	.975	.978	.680	.641
	N	5	5	5	5	5	5	5	5	5	5	5
CSHQ	Pearson Correlation	.717	1	.928*	.351	.210	-.313	-.346	-.092	.508	-.744	-.389
	Sig. (2-tailed)	.173		.023	.562	.734	.608	.568	.884	.382	.150	.517
	N	5	5	5	5	5	5	5	5	5	5	5
Bedtime	Pearson Correlation	.863	.928*	1	.020	.290	-.524	-.669	-.301	.329	-.706	-.185
	Sig. (2-tailed)	.059	.023		.974	.636	.365	.217	.623	.589	.183	.766
	N	5	5	5	5	5	5	5	5	5	5	5
Sleepbehavior	Pearson Correlation	-.228	.351	.020	1	-.117	.206	.678	.293	.244	-.299	-.239
	Sig. (2-tailed)	.712	.562	.974		.851	.740	.208	.632	.692	.625	.699
	N	5	5	5	5	5	5	5	5	5	5	5
waking	Pearson Correlation	-.133	.210	.290	-.117	1	-.765	-.344	-.872	.480	-.784	.490
	Sig. (2-tailed)	.831	.734	.636	.851		.132	.571	.054	.413	.116	.402
	N	5	5	5	5	5	5	5	5	5	5	5
morningwake	Pearson Correlation	-.272	-.313	-.524	.206	-.765	1	.686	.952*	.047	.688	-.717
	Sig. (2-tailed)	.657	.608	.365	.740	.132		.201	.012	.940	.199	.173
	N	5	5	5	5	5	5	5	5	5	5	5
Parentage	Pearson Correlation	-.735	-.346	-.669	.678	-.344	.686	1	.580	.126	.309	-.284
	Sig. (2-tailed)	.157	.568	.217	.208	.571	.201		.305	.840	.612	.644
	N	5	5	5	5	5	5	5	5	5	5	5
Childage	Pearson Correlation	-.019	-.092	-.301	.293	-.872	.952*	.580	1	-.013	.621	-.793
	Sig. (2-tailed)	.975	.884	.623	.632	.054	.012	.305		.983	.264	.110
	N	5	5	5	5	5	5	5	5	5	5	5
Parentconcern	Pearson Correlation	-.017	.508	.329	.244	.480	.047	.126	-.013	1	-.612	-.510
	Sig. (2-tailed)	.978	.382	.589	.692	.413	.940	.840	.983		.272	.380
	N	5	5	5	5	5	5	5	5	5	5	5
Parentsleep	Pearson Correlation	-.254	-.744	-.706	-.299	-.784	.688	.309	.621	-.612	1	-.125
	Sig. (2-tailed)	.680	.150	.183	.625	.116	.199	.612	.264	.272		.841
	N	5	5	5	5	5	5	5	5	5	5	5
childsleepduration	Pearson Correlation	-.286	-.389	-.185	-.239	.490	-.717	-.284	-.793	-.510	-.125	1
	Sig. (2-tailed)	.641	.517	.766	.699	.402	.173	.644	.110	.380	.841	
	N	5	5	5	5	5	5	5	5	5	5	5

*. Correlation is significant at the 0.05 level (2-tailed).

The chiropractic perspective on autism and sleep

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Autistic children present with a multitude of symptoms common to the syndrome. Getting to sleep, maintaining a normal sleep cycle and quality of sleep are often problematic. Many therapeutic approaches, including medications, nutraceutical supplementation, behavioral modification, development of a sleep routine, harmonic resonances, and various forms of bodywork are used with varying degrees of success.[1]

Caregivers of autistic children are known to suffer stress that often contributes to development of disease, reduced resilience, emotional and sleep dysfunction and other markers of poor health.[2] When a parent or guardian on whom the child depends loses capacity to continue to deliver adequate care, both parties suffer and decline.

When individuals are subjected to protracted stress, there is increased sympathetic tone and diminished parasympathetic tone.[3] When an individual is in a prolonged state of fight-or-flight, the stress on the nervous system also reduces immune responses, contributes to chronic inflammation (and, therefore, heart disease, cancer, autoimmunity, diabetes type II and cerebrovascular disorders) and impairs cognitive functions.[4]

There is no single treatment that stands alone in the management of symptoms of autism, rather, combined approaches act synergistically.[5] At Kentuckiana Children's Center, clinicians employ as a foundation of care, the chiropractic adjustment. They have found clinically that when the adjustment is added to the treatment regimen, responses to those therapies improve. Evidence of the enhancing effect of the adjustment is reported subjectively by parents, teachers, other therapists and physicians and extended family members. Parents report improvements in autonomic processes such as sleep.

While the effects of chiropractic adjustments are subject to ongoing research, it is thought that spinal adjustments have a modulating effect on the autonomic nervous system. Mechanism of action of the effects of the adjustment were proposed by Seaman and Winterstein in 1998.[6] These authors conclude that "joint complex dysfunction should be included in the differential diagnosis of pain and visceral symptoms be-cause joint complex dysfunction can often generate symptoms which are similar to those produced by true visceral disease." They demonstrate how dysafferentation may provoke autonomic concomitants that can enhance sympathetic activity. Other studies demonstrate activation of parasympathetic activity and other neuromodulatory effects with spinal manipulative treatment.[7-10]

A common feature among spectrum children is self-injurious behaviors. These may appear has head banging, biting themselves and clawing at skin. Whether these behaviors are uncontrolled tics or whether they may be a reaction to not being understood, or not getting something they want, the behaviors provoke pain and anxiety, which can contribute to sleep disturbances. The adjustment may reduce pain and stress, and therefore, allow for a better night's sleep.[11]

When sleep improves for the autistic child after introducing chiropractic adjustments, not only does that child have reduced stress, but so does the caregiver. Improved quality of sleep contributes to better capacity to perform throughout the day. The caregiver, then, has better quality of life and can provide more effectively for the child.

References:

1 Reynolds A, Malow B. Sleep and autism spectrum disorders. *Pediatr Clin North Am* 2011 Jun;58(3):685-98. doi: 10.1016/j.pcl.2011.03.009.

2 Adelman R, Tmanova L, Delgado D, Dion S, Lachs M. Caregiver burden: a clinical review. *JAMA*. 2014 Mar 12;311(10):1052-60. doi: 10.1001/jama.2014.304.

3 Mulkey S, du Plessis A. Autonomic nervous system development and its' impact on neuropsychiatric outcome. *Pediatr Res*. 2019 Jan; 85(2): 120—126.

4 American Psychological Association. (2018, November 1). Stress effects on the body. https://www.apa.org/topics/stress/body.

5 Hyman S. et al. Identification, Evaluation and management of children with autism spectrum disorder. From the American Academy of Pediatrics, *Clinical Report* January 1, 2020.

6 Seaman D, Winterstein J. Dysafferentation: A novel term to describe the neuropath-ophysiological effects of joint complex dysfunction. *J Manipulative Physiol Ther* (May); 21 (4): 267-280.

7 Valenzuela PL, Pancorbo S, Lucia A, Germain F. Spinal manipulative therapy effects in autonomic regulation and exercise performance in recreational healthy athletes: A randomized controlled trial *Spine*. 2019 May 1;44(9):609-614. doi: 10.1097/BRS.0000000000002908.

8 Taylor H, Holt K, Murphy B. Exploring the neuromodulatory effects of the vertebral subluxation and chiropractic care. *Chiropr J Aust* 2010;40:37-44.

9 Pickar JG. Neurophysiological effects of spinal manipulation. *Spine J.* 2002;2(5):357-71.

10 Haldeman S. Neurologic effects of the adjustment. *J Manipulative Physiol Ther*. 2000;23(2):112-4.

11 Dagenais S, Haldeman S. Chiropractic. *Prim Care*. 2002 Jun;29(2):419-37. doi: 10.1016/s0095-4543(01)00005-7.

Physiology of sleep disturbances in ASD and naturopathic treatment strategies

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Introduction

In recent years the emerging study of sleep science has established that sleep is one of the cornerstones of health in adults and children. Poor sleep quality and quantity can have deleterious effects on child development in the following areas: physical growth, immune function, cognitive development, behavior, emotional regulation, attention, memory, learning, speech and language development, and sensory integration. Poor sleep predisposes adults to myriad negative health outcomes as well, such as depression, hypertension, type 2 diabetes, asthma, allergy, and cardiovascular disease.[1] Sleep problems are reported in up to 80% of children and up to 50% of adults with Autism Spectrum Disorders (ASD) making this an issue that affects the ASD population and their caregivers across the lifespan.[2] The author of the main manuscript conducted a small study, which revealed that improvement in sleep quality and quantity has positive outcomes for both patient and caregiver. The purpose of this paper is to review common causes of sleep disturbance in ASD and highlight naturopathic treatment options for improved sleep outcomes.

Physiology of Sleep disturbances in ASD

Sleep disturbances fall into two categories: Insomnia and Circadian Sleep-Wake disorder. Insomnia is defined as the difficulty falling or staying asleep. Circadian Sleep-Wake disorder is defined by difficulty aligning the timing of the sleep wake cycle.[3] Sleep disorders are regulated by exogenous processes like sleep environment, sleep hygiene, and routine, or endogenous processes like hormone and neurotransmitters production and regulation, and the gut microbiome.[3] Successful interventions depend on determining which process, or combination of processes are implicated in each person's sleep disorder. This person-centered approach requires thorough biomedical assessment and patient specific intervention.

Sleep Hygiene and Environmental Factors

Establishing successful sleep habits can be challenging for the ASD population but is essential in obtaining adequate sleep for children and caregivers. Sleep hygiene has an important role in sleep quality and quantity, especially when established early in childhood.[4] Establishing a regular bedtime routine, even on weekends, prohibiting television in the bedroom, and eliminating caffeinated drinks are important tenets of sleep hygiene.[4] It is also important for parents of children with ASD to closely monitor and curb access to electronic media devices such as tablets, smart phones, and computers. Access to these devices, especially in the bedroom, results in shorter sleep duration and daytime sleepiness when compared with those who did not have bedroom access.[5] Studies show that children with ASD spend more time playing video games rising to the level of "problematic" video game use than their non-ASD peers.[5]

Neurotransmitters

Transition from the sleep-wake cycle is regulated by a complex interplay between neurotransmitters and hormones. Sleep promotion is governed by gaba-aminobutyric acid (GABA) and melatonin. Arousal and wakefulness are governed by serotonin, acetylcholine and glutamate.

GABA is an inhibitory neurotransmitter important for transition into sleep. A recent study found a reduction in GABA-A receptors in people with ASD caused disruption in sleep-wake behaviors.[6] Perhaps as important as adequate levels of GABA and function of the GABA receptors, is the balance between GABA's inhibitory effect and glutamate's excitatory effect. These two neurotransmitters act in tandem to function as a "switch" to transition between sleep and wakefulness, so balance here is key for optimum sleep.[7]

Melatonin is the other sleep promoting neurotransmitter. Melatonin is produced in the pineal gland in response to light/dark exposure. Disruptions in metabolism and/or production of melatonin are common in the ASD population and are implicated in sleep disturbances.[8] Other studies show that some individuals with ASD produce lower levels of melatonin than typical night-time levels causing delayed transition into sleep.[9] Exposure to blue light from electronics, especially in the hours before bedtime, suppresses melatonin and can further contribute to sleep disturbances.[10]

Acetylcholine is necessary for rapid eye movement (REM) sleep which is highly active in the developing brain. Acetylcholine is markedly reduced in ASD.[11] In a small study, it was concluded that children benefitted from improving acetylcholine levels by inhibiting acetylcholinesterase, which resulted in improved outcomes in REM sleep, behavior, and attention.[12]

Cortisol Dysregulation in ASD

Many children with ASD have a dysfunctional hypothalamic-pituitary-adrenal (HPA) axis contributing to abnormal cortisol patterns. The HPA axis releases cortisol from the adrenal cortices in response to stress. Typically,

cortisol is released in a binaural fashion. It is highest in the morning, decreasing throughout the day and at its lowest level in the evening. Studies have shown that children with ASD have higher evening cortisol[13] and lower morning cortisol levels[14] contributing to blunted diurnal slope in comparison to neurotypical children.[13] The research has shown that there is no difference in cortisol output between neurotypical children and children with ASD. This suggests that HPA axis dysfunction may be due to an accumulation of stress throughout the day, which leads to the increased evening cortisol levels seen in children with ASD.[14] In addition, increased cortisol response in children with ASD has been associated with increased stress response, sensory sensitivity, and anxiety.[15] Abnormal cortisol levels due to dysfunctional HPA axis contributes to various symptoms associated with the ASD diagnosis, such as sleep disturbances, anxiety, sensory overload, hyperactivity, OCD, perseverations, aggression, weight issues, etc.

Gut Microbiota and Sleep in ASD

Many children with ASD have imbalances in their gut microbiome. Candida overgrowth, parasitic infections, and bacterial imbalances in the gut microbiome can be found in children with ASD. Differences in sensory patterns resulting in restricted eating, altered ability to fend off gut infections, and increased intestinal permeability are all common in the ASD population.[16] Emerging research demonstrates the necessity of a healthy gut for the production of serotonin and its effect on the gut-brain axis. For example, sleep disturbances that occur around the lunar cycle may be due to parasitic activity based on our body's natural fluctuation in serotonin and melatonin levels. Around the full moon, more serotonin is produced and parasites respond to elevated serotonin production with increased activity.[17] In addition, candida albicans may increase production of serotonin in the intestines at the expense of decreasing serotonin production in the brain.[18] Therefore, alterations in gut microbiome may negatively affect serotonin production and metabolism and therefore impact sleep regulation in those with ASD.

Seizures and Sleep in ASD

It is estimated that 2/3 of children with ASD will have abnormal EEG findings. 1/3 will be diagnosed with epilepsy and demonstrate typical signs of seizure activity. The other 1/3 may have subclinical EEG changes with no outward signs of seizure activity.[19] Abnormal electrical discharge and seizures can negatively affect sleep and should be assessed in children with an ASD diagnosis. Given that a third of children with autism do not have a diagnosis of epilepsy, but are experiencing subclinical EEG changes, this underlying cause of sleep disturbance often goes undiagnosed. Causes of abnormal EEG findings in the ASD population can include metabolic disturbances, mitochondrial disorders, cerebral folate deficiency, inflammation, and genetic disorders.[20] Sleep disorders and epilepsy (abnormal neural discharges) have a reciprocal relationship. Disordered sleep, common in ASD, predisposes this population to abnormal neural discharges, and conversely abnormal neural discharges can cause or exacerbate sleep disorders.[21] Therefore, if a child with ASD is experiencing sleep disturbances it is important to rule out seizure activity.

Naturopathic Interventions for Sleep in ASD

While most pediatricians recognize the prevalence of sleep disorders in the ASD population, after sleep hygiene is addressed, pharmacologic intervention is the next step of intervention.[22] Naturopathic interventions provide safe and effective alternatives to pharmacologic solutions for those whom medications were not successful and/or those looking for natural alternatives to prescription interventions. After extensive biomedical testing to determine functional levels of gut dysbiosis, neurotransmitters, hormones, and nutritional status, appropriate supplementation can restore homeostasis.

Practitioners can balance neurotransmitter levels directly by appropriate supplementation. Levels can also be altered by supplementation of cofactors and food sources that support the body's ability to produce neurotransmitters naturally. GABA requires B6 for its synthesis so ensuring adequate levels of B6 is an important consideration for treatment.[23] Vitamin D, magnesium, and zinc should also be considered to support optimal GABA levels.[24] Lactobacillus plantarum and Lactobacillus brevis are helpful in reestablishing healthy gut flora and have also been found to increase the conversion of GABA from glutamate.[25] In this way Lactobacillus can act to indirectly support sleep via two mechanisms. Food sources of GABA include fermented soy and milk products, although children with food sensitives or allergies to milk or soy should continue to eliminate them from their diets.[26] Exercise has also been shown to increase GABA levels and should be included as a treatment strategy to support sleep.[27]

Melatonin is synthesized from serotonin and both play an important role in the sleep-wake cycle. This biochemical pathway, starting with tryptophan, requires multiple B vitamins including folate and Vitamin B12, as well as, zinc, and magnesium. A well-balanced diet supports melatonin production especially when diet is focused on vegetables, fruits, and grains.[28] Perhaps the most important way to support melatonin production is decreasing exposure to light sources before bed.[28] Removing electronic devices like tablets, computers, smart phones from the bedroom and limiting their use in the hours before bedtime should be an important part of the treatment of sleep disturbance in ASD. Decreasing electronic use can be very beneficial for regulating cortisol. In addition to decreasing screen time, supporting one's adrenal glands with the use of adequate hydration, salt, and herbal adaptogens may be necessary to regulate cortisol patterns in children with ASD. Ashwagandha root (Withania somnifera)

is an herbal that can improve sleep quality and sleep onset in those suffering from insomnia.[29]

Adequate levels of acetylcholine are required for restorative REM sleep. Vitamin B5 (Pantothenic acid) is required to produce acetylcholine in the body, so this should be considered in treatment of sleep in ASD. Adequate levels of vitamin B12 are also helpful in optimizing this neurotransmitter, as well as dietary sources of choline such as fish, eggs, beef, soybeans, and chicken breast.[30]

Addressing gut dysbiosis is an important part of treating sleep in ASD as many children with ASD suffer from some form of dysbiosis. Testing to determine levels of pathogenic microbiota and accurately assessing imbalances should be done at the initiation of treatment. This information can then be used to establish an individualized treatment plan to establish healthy flora. For yeast overgrowth, antimicrobials such as grapefruitseed extract, pau d'arco, and gynmena slyvestre could be considered. Grapefruitseed extract inhibits the growth of candida biofilms.[31] Pau D'arco inhibits the biochemical process that fungi need to produce oxygen and energy to survive.[32] Gymenma Slyverstre blocks the virulence properties of fungus by inhibiting the hyphal growth of Candida albicans.[33] In addition, Saccromyces Boulardii, which has prebiotic and probiotic properties, decreases candida ability to form filaments, adherence to the intestine, and biofilm formation.[34] For anti-parasitic support, treatments may include antimicrobials herbals such as artemisia and black walnut. Artemisia contains artemisinin, which has potent anti-malarial and anti-parasitic properties.[35] A major constituent in Black Walnut, known as Juglone, inhibits enzymes necessary for parasites metabolic function.[36] Therefore, antimicrobial herbals against the specific germ can be effective at reestablishing a healthy microbiome and improve symptoms.

For those children with ASD who have abnormal EEG activity as a causal factor, identifying the underlying cause is paramount. Once causal factors are identified, person-centered treatment can be utilized to remediate specific processes. In general, people with abnormal EEG activity benefit from adopting a ketogenic diet given its ability to calm neuroinflammation.[37] Nutritional interventions may be beneficial in treating epileptic encephalopathies including Taurine, Vitamin B6, Magnesium, Omega 3 fatty acids, L-Carnosine, and Folinic acid.[38] Identifying and treating cerebral folate deficiency with Folinic acid can help remediate metabolic causes of sleep disturbances in ASD.[39]

Further attention should be given to nutritional deficiencies of iron and magnesium levels in children with ASD experiencing sleep disturbances. Given the restrictive diets of many children with ASD, nutrient deficiencies are common. Research shows that while many children with ASD have lower hematocrit, hemoglobin, and iron levels the levels may not result in anemia.[40] Ferritin levels less than 50 ng/ml have been associated with Restless Leg Syndrome and Periodic Limb Movement Disorder in children.[41] Given that iron deficiency and ferritin levels less than 50 ng/ml are associated with sleep disturbances, it is prudent to assess and remediate iron deficiency in those with ASD and sleep disturbances.[42] It is important to implement iron under the guidance of one's physician due to the potential of iron complicating underlying gut dysbiosis inadvertently by "feeding" pathogenic bacteria that thrive on iron (and reduce available iron for absorption via the gut).[43] Decreased magnesium status has been associated with ASD and sleep disturbances. Supplementing magnesium has been found to improve sleep quality regardless of cause.[44] Magnesium is a cofactor for the production of GABA, and this may explain the improved sleep quality and quantity noted with supplementation of magnesium. Magnesium also antagonizes the NMDA (glutamate) receptor so optimizing magnesium levels will help balance GABA/glutamate levels needed to regulate the sleep-wake cycle.

Conclusion

Sleep disorders are prevalent in 80% of children and 50% of adults with ASD. Sleep disorders, although very common in ASD, have myriad causes. Using a person-centered approach to identify and treat specific causes can provide significant improvement in sleep outcomes. Naturopathic interventions provide powerful ways to remediate imbalances and can have profound effect in reducing or eliminating sleep disorders, improving the quality of life for the individuals with ASD and their caretakers. Interventions explored in this paper include balancing important neurotransmitters and hormones, treating gut dysbiosis, assessing nutritional deficiencies, dietary interventions, and establishing good sleep hygiene. Neurological workup should be included to rule out seizure and subclinical seizure activity as a causal factor for sleep disorders. In addition to topics discussed above, mitochondrial function, food sensitivities, allergies, enlarged tonsils/adenoids, constipation, medication reactions, cerebral folate deficiency, blood sugar regulation, and identification of genetic SNPs should all be considered as part of a thorough treatment plan.

References:

1. Bollu PC, Kaur H. Sleep medicine: Insomnia and sleep. *Mo Med*. 2019;116(1):68-75.

2. Ballester P, Richdale AL, Baker EK, Peiró AM. Sleep in autism: A biomolecular approach to aetiology and treatment. *Sleep Med Rev*. 2020;54:101357.

3. Matenchuk BA, Mandhane PJ, Kozyrskyj AL. Sleep, circadian rhythm, and gut microbiota. *Sleep Med Rev*. 2020;53:101340.

4. Bathory E, Tomopoulos S. Sleep regulation, physiology and development, sleep duration and patterns, and sleep hygiene in Infants, toddlers, and preschool-age children. *Curr Probl Pediatr Adolesc Health Care*. 2017;47(2):29-42.

5. Mazurek MO, Engelhardt CR, Hilgard J, Sohl K. Bedtime Electronic Media Use and Sleep in Children with Autism Spectrum Disorder. *J Dev Behav Pediatr.* 2016;37(7):525-531.

6. Mesbah-Oskui L, Penna A, Orser BA, Horner RL. Reduced expression of a5GABAA receptors elicits autism-like alterations in EEG patterns and sleep-wake behavior. *Neurotoxicol Teratol*. 2017;61:115-122.

7. Schneider L. Neurobiology and Neuroprotective Benefits of Sleep. *Continuum* (Minneap Minn). 2020;26(4):848-870.

8. Rossignol DA, Frye RE. Melatonin in autism spectrum disorders: a systematic review and meta-analysis. *Dev Med Child Neurol*. 2011;53(9):783-792.

9. Goldman, S.E., Adkins, K.W., Calcutt, M.W. et al. Melatonin in Children with Autism Spectrum Disorders: Endogenous and Pharmacokinetic Profiles in Relation to Sleep. *J Autism Dev Disord* 44, 2525—2535 (2014).

10. West KE, Jablonski MR, Warfield B, et al. Blue light from light-emitting diodes elicits a dose-dependent suppression of melatonin in humans. *J Appl Physiol* (1985). 2011;110(3):619-626.

11. Omura Y, Lu D, Jones MK, et al. Early Detection of Autism (ASD) by a Non-invasive Quick Measurement of Markedly Reduced Acetylcholine & DHEA and Increased ß-Amyloid (1-42), Asbestos (Chrysotile), Titanium Dioxide, Al, Hg & often Coexisting Virus Infections (CMV, HPV 16 and 18), Bacterial Infections etc. in the Brain and Corresponding Safe Individualized Effective Treatment. *Acupunct Electrother Res*. 2015;40(3):157-187.

12. Ballester P, Richdale AL, Baker EK, Peiró AM. Sleep in autism: A biomolecular approach to aetiology and treatment. *Sleep Med Rev*. 2020;54:101357.

13. Muscatello R & Corbett BA (2018). Comparing the Effects of Age, Pubertal Development, and Symptoms Profile on Cortisol Rhythm in Children and Adolescents with Autism Spectrum Disorder. *Autism Research*, 11, 110 -120.

14. Corbett BA, Schupp CW, Levine S & Mendoza S (2009). Comparing cortisol, stress, and sensory sensitivity in children with autism. *Autism Research*, 2, 39-49.

15. Corbett BA, Mendoza S, Abdullah M, Wegelin JA & Levine S (2006). Cortisol circadian rhythms and response to stress in children with autism. *Psychoneuroendocrinology*,31, 59-68.

16. Ristori MV, Quagliariello A, Reddel S, et al. Autism, Gastrointestinal Symptoms and Modulation of Gut Microbiota by Nutritional Interventions. *Nutrients*. 2019;11(11):2812.

17. Bagnaresi P et al. (2012) The role of melatonin in parasite biology. *Molecular and Biochemical Parasitology*, 181, 1-6.

18. Srikantha P, Mohajeri MH. The possible role of the microbiota-gut-brain-axis in autism spectrum disorder. *Int. J. Mol. Sci*. 2019;20:2115

19. Spence S, Schneider M. The Role of Epilepsy and Epileptiform EEGs in Autism Spectrum Disorders. *Pediatr Res*. 2009;65: 599-606.

20. Frye RE. Metabolic and mitochondrial disorders associated with epilepsy in children with autism spectrum disorder. *Epilepsy Behav.* 2015;47:147-157.

21. Malow BA. Sleep disorders, epilepsy, and autism. *Ment Retard Dev Disabil Res Rev*. 2004;10(2):122-125.

22. van der Heijden KB, Stoffelsen RJ, Popma A, Swaab H. Sleep, chronotype, and sleep hygiene in children with attention-deficit/hyperactivity disorder, autism spectrum disorder, and controls. *Eur Child Adolesc Psychiatry*. 2018;27(1):99-111.

23. Jewett BE, Sharma S. Physiology, GABA. FL: StatPearls Publishing;2021.

24. Mills DJ. The Aging GABAergic System and Its Nutritional Support. *J Nutr Metab*. 2021;2021:6655064.

25. Yunes RA, Poluektova EU, Vasileva EV et al. A Multi-strain Potential Probiotic Formulation of GABA-Producing Lactobacillus plantarum 90sk and Bifidobacterium adolescentis 150 with Antidepressant Effects. *Probiotics & Antimicro. Prot*. 2020;12:973-979.

26. Oketch-Rabah HA, Madden EF, Roe AL, Betz JM. United States Pharmacopeia (USP) Safety Review of Gamma-Aminobutyric Acid (GABA). *Nutrients*. 2021;13(8):2742.

27. Kramer JM, Beatty JA, Plowey ED, Waldrop TG. Exercise and hypertension: a model for central neural plasticity. *Clin Exp Pharmacol Physiol*. 2002;29(1-2):122-126.

28. Peuhkuri K, Sihvola N, Korpela R. Dietary factors and fluctuating levels of melatonin. *Food Nutr Res*. 2012;56:10.

29. Langade D, Kanchi S, Salve J, Debnath K, Ambegaokar D. Efficacy and Safety of Ashwagandha (Withania somnifera) Root Extract in Insomnia and Anxiety: A Double-blind, Randomized, Placebo-controlled Study. *Cureus*. 2019;11(9):e5797.

30. Hollenbeck CB. An introduction to the nutrition and metabolism of choline. *Cent Nerv Syst Agents Med Chem*. 2012;12(2):100-113.

31. Tsutsumi-Arai C, Takakusaki K, Arai Y, Terada-Ito C, Takebe Y, Imamura T, Ide S, Tatehara S, Tokuyama-Toda R, Wakabayashi N, Satomura K. Grapefruit seed extract effectively inhibits the Candida albicans biofilms development on polymethyl methacrylate denture-base resin. *PLoS One*. 2019;14(5).

32. Portillo A, Vila R, Freixa B, Adzet T, Canigueral S. Antifungal activity of Paraguayan plants used in traditional medicine. *J Ethnopharmacol*. 2001;76:93-98.

33. Vediyappan G, Dumontet V, Pelissier F, d'Enfert C. Gymnemic acids inhibit hyphal growth and virulence in Candida albicans. *PLoS One*. 2013 Sep 11;8(9).

34. Tomicic ZM. Beneficial properties of probiotic yeast Saccharomyces boulardii. *Food Feed Res*. 2016;43(2):103-110.

35. Krishna S, Bustamante L, Haynes RK, Staines HM. Artemisinins: their growing importance in medicine. *Trends Pharmacol Sci*. 2008 Oct;29(10):520-7.

36. Jha BK, Jung HJ, Seo I, Suh SI, Suh MH, Baek WK. Juglone induces cell death of Acanthamoeba through increased production of reactive oxygen species. *Exp Parasitol*. 2015;159:100-6.

37. Koh S, Dupuis N, Auvin S. Ketogenic diet and Neuroinflammation. *Epilepsy Res*. 2020;167:106454.

38. Agadi S, Quach M, Haneef Z. Vitamin-Responsive Epileptic Encephalopathies in Children. *Epilepsy Research and Treatment*. 2013; 1-8.

39. Rossignol DA, Frye RE. Cerebral Folate Deficiency, Folate Receptor Alpha Autoantibodies and Leucovorin (Folinic Acid) Treatment in Autism Spectrum Disorders: A Systematic Review and Meta-Analysis [published correction appears in J Pers Med. 2022 Apr 29;12(5):]. *J Pers Med*. 2021;11(11):1141.

40. Gunes S, Ekinci O, Celik T. Iron deficiency parameters in autism spectrum disorder: clinical correlates and associated factors. *Ital J Pediatr.* 2017;43(1):86.

41. Donskoy I, Loghmanee D. Iron and Insomnia in Autism Spectrum Disorder. *Pediatr Neurol Briefs*. 2020 Dec 9;34:17.

42. Leung W, Singh I, McWilliams S, Stockler S, Ipsiroglu OS. Iron deficiency and sleep - A scoping review. *Sleep Med Rev*. 2020;51:101274.

43. Seyoum Y, Baye K, Humblot C. Iron homeostasis in host and gut bacteria - a complex interrelationship. *Gut Microbes*. 2021 Jan-Dec;13(1):1-19.

44. Skalny AV, Mazaletskaya AL, Ajsuvakova OP, et al. Magnesium Status in Children with Attention-Deficit/Hyperactivity Disorder and/or Autism Spectrum Disorder. *Soa Chongsonyon Chongsin Uihak*. 2020;31(1):41-45.

How sensory integration disorder can contribute to sleep disturbances in autistic children

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Children with Autism Spectrum Disorder (ASD) are at an increased risk for sleep disturbances, and studies indicate that between 50 and 80% of children diagnosed on the autistic spectrum experience sleep problems.[1] One area that is challenging for many autistic children is their ability to receive and interpret sensory stimuli. This author will focus on the impact that dysregulation of the sensory processing system may have on sleep disturbances in this population and interventions to assist with proper sleep.

Statistics published in the *Journal of Pediatric Neuroscience* in 2015 reported the following instances of sleep issues in autistic children:

- 54% displayed resistance to bedtime
- 56% experienced insomnia
- 53% suffered from parasomnias, such as sleepwalking or night terrors
- 25% experienced sleep disordered breathing, including sleep apnea
- 45% had difficulty waking up in the morning
- 31% experienced daytime sleepiness[2]

This year (2022) in a scoping review, Lane et al wrote that "the incidence of sensory reactivity differences in autism exceeds that in the neurotypical population. The basis of sleep disorders in autism is multifactorial, but sensory integration/processing concerns may play a role. Research that investigates this interplay for autistic individuals is limited but vital."[3]

A study in the *Journal of Molecular Autism* examined the relationship between sleep disturbances and sensory sensitivities in autistic children. This study found a correlation between tactile, visual, auditory, and oral sensitivities and sleep disturbances in children with autism. Tactile hypersensitivity was found to be the most frequent cause of sleep disturbance scoring 25%.[4]

The study from the *Journal of Pediatric Neuroscience* gives a framework for the varied presentations of sleep disturbances experienced by autistic individuals. The reasons for these disrupted sleep patterns are varied and are most likely caused by neurological, physiological, and environmental factors.[2] People with autism will have varied responses to stimuli but most are classified into two categories; hypersensitivity and hyposensitivity. A child with hypersensitivity is described to have a heightened sensitivity to stimulation. For example, accidentally bumping into a child may not bother a child with a typical neurological response however this same bumping into could be very overwhelming and disruptive to a child with tactile sensitivity. Therefore, a child going to sleep may be overwhelmed by the feel of their pajamas or sheets or a sibling in the same bed and this may impact their ability to sleep. The child with hyposensitivity to tactile input may need a weighted blanket or compression blanket to stimulate the deep joint mechanoreceptors and help them regulate their autonomic nervous system to fall asleep. Each of the senses including the tactile, auditory, vestibular, visual, olfactory, and proprioceptive have input to the sensory system. Any or all of this input can be misinterpreted ("jumbled up", heightened (sensory overload) or dampened (sensory deprivation) and any dysfunction or alteration in "reception" can result in sleep disturbances.

Interventions should be aimed at helping the person to normalize neurological responses to stimuli, help regulate bodily functions and create an environment that supports sleep.[3,5]

Interventions

Qi Gong Sensory Treatment

In Chinese Medicine autism is characterized by a block of one or more of the sensory pathways and stimuli cannot be properly received and processed. "The sensitivity of one or more of the sensory channels disturbs the normal flow of Qi, causing deficiencies or accumulations inside the head. These phenomena cause the many different hyper- and hypo-sensitivities that autistic children show in response to touch, pain, noise, taste, olfactory and visual stimuli."[6] Qi Gong massage aims to clear the accumulations and strengthen the deficiencies of Qi the child is exhibiting. This treatment is taught to parents and supported with treatment from a Qi Gong Sensory Treatment (QST) therapist for at least five months. The person giving the massage uses their hands to provide a tapping (clearing) or pressing (deep pressure) touch down the acupuncture meridians. The person giving the massage monitors the child's responses and will change the movement to help either move an energy blockage or to fill the emptiness where the energy is not flowing.

"Research shows that pleasure and bonding with gentle touch are mediated by tiny sensory nerves in the skin, and when these nerves are damaged, children lose pleasure and bonding with touch. Fortunately, the damage is reversible, and treatment with QST for autism returns the sense of touch to normal. This improves bonding, stimulates social development, and eliminates autistic behavior."[7,8]

Cranial Sacral Therapy

Cranial Sacral Therapy[9] is a light touch therapy that aims

to balance the craniosacral system. The cranial system is comprised of fluid that surrounds and cushions the brain and spinal cord, providing nutrients and removing waste material. Treatment of this system uses light touch to release bone and membranous restrictions. This treatment helps to balance the autonomic nervous system.

The focus of CST is to enhance the movement of:

- The three fascial membrane layers surrounding the brain.
- The fluid (blood and cerebrospinal fluid) moving through the vessels (blood/lymph) of the cranium and throughout tissues of the central nervous system (brain and spinal cord)
- Address adhesions, restriction or tension in areas of the body that are adjacent to the craniosacral system causing fascial strain that inhibits the normal cranial sacral rhythm within the system.

This light work promotes improved self-regulation of the autonomic nervous system therefore helping to normalize many physiologic functions overall. Cranial sacral therapy is a gentle but clinically effective modality. While gentle, practicing craniosacral therapy requires advanced training as it involves very specific application of very gentle techniques which, when applied correctly, can produce very significant results.[10]

Occupational Therapy — Ayers Sensory Integration

Although it is beyond the scope of this article, the Ayres Sensory Integration intervention is one of the most frequently requested and highly utilized interventions in autism from the occupational therapists. This intervention has specific requirements for therapist qualifications and the process of therapy. A systematic review done in 2015 requesting further rigorous research was needed was followed up in 2019 with another systematic review of studies providing Ayres Sensory Integration therapy to children with autism indicating that it is an evidence-based practice according to the criteria of the Council for Exceptional Children and is another avenue for parents to explore.[11,12]

Strategies for a better bedtime at home

Strategies to help the over responsive child to self-regulate and soothe at bedtime:

Environment: Bedtime should begin at least an hour before sleep. Have a routine that is followed nightly and supports self-regulation. Reading books, quiet play, low lighting, no screens and avoid giving any fruit juice or sugary treats or desserts (even high sugar fruits, like grapes and cherries could raise their blood sugar!).

Clothing: Allow the child to choose what feels comfortable to sleep in. If they need to sleep without clothes, let them sleep without clothes and keep the room comfortably warm. Children with tactile sensitivities often are overwhelmed by clothing. Pay attention to the fabrics they prefer and provide them choices for bedtime.

Sheets and blankets: Try different sheet fibers and tensions. An autistic child might prefer a weighted blanket over as standard blanket, or they may choose to use no blanket at all. There are even compression blankets made of lycra that fit over the mattress and provide deep joint compression while in bed. They are also useful in controlling excessive movement and falling out of bed without preventing all movement.[13-15]

In conclusion, therapeutic interventions may be helpful in ameliorating the many sequelae of poor sleep (the child and the family). It should also not be overlooked that autistic children are children, subject to the same social and emotional influences that disrupt the sleep of neurotypical children and the adults that caretake them. The issues are sometimes best addressed by working with the family unit as well as the autistic child.

References:

1.Mazzone L, Postorino V, Siracusano M, Riccioni A, Curatolo P. The Relationship between Sleep Problems, Neurobiological Alterations, Core Symptoms of Autism Spectrum Disorder, and Psychiatric Comorbidities. *J Clin Med*. 2018 May 3;7(5):102. doi: 10.3390/jcm7050102. PMID: 29751511; PMCID: PMC5977141.

2. Devnani PA, Hegde AU. Autism and sleep disorders. *J Pediatr Neurosci*. 2015 Oct-Dec;10(4):304-7. doi: 10.4103/1817-1745.174438. PMID: 26962332; PMCID: PMC4770638.

3. Lane SJ, Leão MA, Spielmann V. Sleep, Sensory Integration/Processing, and Autism: A Scoping Review. *Front Psychol*. 2022 May 17;13:877527. doi: 10.3389/fpsyg.2022.877527. PMID: 35656493; PMCID: PMC9152214. https://www.frontiersin.org/articles/10.3389/fpsyg.2022.877527/full.

4. Tzischinsky O, Meiri G, Manelis L, Bar-Sinai A, Flusser H, Michaelovski A, Zivan O, Ilan M, Faroy M, Menashe I, Dinstein I. Sleep disturbances are associated with specific sensory sensitivities in children with autism. *Mol Autism*. 2018 Mar 27;9:22. doi: 10.1186/s13229-018-0206-8. PMID: 29610657; PMCID: PMC5872526.

5. Quake-Rapp C, Atchison B. Sensory processing disorders and treatment: Occupational therapy using a sensory integration approach. In *Behavioral Pediatrics*: Fourth Edition, Nova Science Publishers, Inc. 2015; pp. 165-180.

6. Silva L, Schalock M, Gabrielsen K, Budden S, Buenrostro M, and Horton G. (2015). *Autism Research and Treatment*, Volume 2015 (2015), Article ID 904585.

7. Silva LM, Cignolini A. A medical qigong methodology for early intervention in autism spectrum disorder: a case series. *Am J Chin Med*. 2005;33(2):315-27. doi: 10.1142/S0192415X05002837. PMID: 15974490.

8. Silva LM, Schalock M, Gabrielsen KR, Hd G. One- And Two-year Outcomes of Treating Preschool Children with Autism with a Qigong Massage Protocol: An Observational Follow-along Study. *Alternative & Integrative Medicine*, 2016, 1-10. https://www.hilarispublisher.com/open-access/one-and-twoyear-outcomes-of-treating-preschool-children-with-autism-with-a-qigong-massage-protocol-an-observational-followalong-st-2327-5162-1000216.pdf.

9. Vredevoogd JD, Upledger JE. *Craniosacral Therapy,* Eastland Press, 1983. ISBN 10: 0939616017 / ISBN 13: 9780939616015.

10. Wanveer T, Upledger J. Autsim Spectrum Disorder: How Cranial Sacral Therapy can help. *Massage Today*, May 2009. Retrieved on Oct14,2022: https://www.massagetoday.com/articles/13656/Autism-Spectrum-Disorder-How-CranioSacral-Therapy-Can-Help.

11. Case-Smith J, Weaver LL, Fristad MA. A systematic review of sensory processing interventions for children with autism spectrum disorders. *Autism*. 2015 Feb;19(2):133-48. doi: 10.1177/1362361313517762. Epub 2014 Jan 29. PMID: 24477447.

12. Schoen SA, Lane SJ, Mailloux Z, May-Benson T, Parham LD, Smith Roley S, Schaaf RC. A systematic review of ayres sensory integration intervention for children with autism. *Autism Res*. 2019 Jan;12(1):6-19. doi: 10.1002/aur.2046. Epub 2018 Dec 12. PMID: 30548827; PMCID: PMC6590432.

13. Chaves J. The Relationship between Sensory Processing and Sleep. Published online by the Center for Connection April 2016. Retrieved on Oct 14, 2022: https://www.thecenterforconnection.org/blogarchive/2016/4/27/the-relationship-between-sensory-processing-and-sleep.

14. The Sleep Help Institute. Sleep Help for Those Diagnosed With ASD. Published online by Living Autism and retrieved on Oct 14, 2022: https://livingautism.com/sleep-help-for-those-diagnosed-with-asd/.

15. Magee L, Goldsmith LP, Chaudhry UAR, et al. Nonpharmacological Interventions to Lengthen Sleep Duration in Healthy Children: A Systematic Review and Meta-analysis. *JAMA Pediatr*. Published online September 12, 2022. doi:10.1001/jamapediatrics.2022.3172.

The dentist's role in the treatment of autistic children and sleep disordered breathing

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Editor's note: *Having conducted an interview with Dr. Leonard Kundel about children on the autistic spectrum and reasons for poor sleep, there was agreement on the importance of emphasizing that the ever growing number of children diagnosed on the autistic spectrum were children first and foremost. This was also true of children with any non-neuro-typical, genetic or traumatically induced physical, mental, chemical or intellectual disabilities. They are all children first. And, as is true for all humans, big and small, there is the frequent unfortunate occurrence of comorbidities. A wholistic approach evaluating children is the most effective route to discern the root causes of a problem and what treatment protocol will best ameliorate some, if not in some cases, all of their symptoms as was documented by a mother of a young man with behavioral issues in the moving video entitled Finding Connor Deegan.*[1] *This last commentary is a segue to Dr. Cantwell's manuscript, "The dentist's role in the treatment of sleep disordered breathing."*

Breathing disordered sleep or sleep disordered breathing - which comes first, the chicken or the egg? Is sleep so poor that breathing is compromised or is one sleeping poorly because breathing is compromised? Both sides of the coin are valid. If the body must struggle to move air in and out, whether it's due to a pulmonary disorder, thoracic spine/rib restriction or an obstructed airway due to enlarged tonsils and adenoids, chronic inflammation (allergies), hyoid displacement or a malformed palate or a tongue tie or tongue size,[2-4] then essentially "nothing will be right." How can it be? If someone is drowning, no amount of psychotherapy nor nutritional supplements will save them. They need a life vest! If breathing becomes easier, the body can function normally. There is an inherent wisdom in the body which is about checks and balances: what is okay and what is not okay? If there is even a small amount of difficulty in obtaining oxygen, behavior will change based on the adaptive capacity – some people can handle the challenges and adapt readily and some fall into distress much more quickly.[5]

The skeletal structures of children since industrialization have become less well developed. The cranium and orofacial structures are not growing ideally lacking the stimulus of early introduction to "hard" textured foods which triggers chewing, lateralizing food between the molars with the tongue and swallowing. Proper swallow becomes compromised early in life when parents prematurely introduce spoons, usually at around six months, when the throat anatomy is not yet mature enough to accommodate an adult-like swallow. This continues with the introduction of nourishment through sucking conduits such as bottles, sippy cups, straws, and food pouches. When a problem is noted, the child is prescribed feeding, speech or myofunctional therapy and their aberrant swallow is overlooked and remains undiagnosed, therefore never addressed and corrected. Proper swallow provides a coherent message to the brain guiding growth and development. In the author's opinion, children of the last several generations appear to be suffering from "the disease of the aristocracy." Parents strive to do everything for their children in the name of good parenting – children have little need to struggle to survive. Foods are soft and less nutritious. Muscles do not load on bones, therefore bones do not respond to the stimulus and grow optimally. Children are attached to pacifiers, are thumb suckers, tongue thrusters, mouth breathers and are generally unhealthy (plagued by allergies, chronic ear infections and sinusitis, gastrointestinal issues and obesity, to name a few health concerns). Tongue and other muscles of mastication must provide a coherent message to the brain and then to the jaws and surrounding bones (directly or indirectly) how to grow in size, density, position and shape.[6]

Parents who pay attention to nutrition often observe that their child's overall development improves with better nutrition. This may be related to the quality of the food (minerals and other nutrients), but also to the density and texture of the foods. "Baby Led Weaning" has been popularized to return to the act of masticating hard foods, avoiding infantile oral behaviors and promoting proper oral motor skills and craniofacial development.[7] There is no one ideal approach but this offers a combination of optimizing function (masticating/swallowing) and nutrition (healthy food; bountiful nutrients/minerals). Children wind up having deficient airways because of poor osseous structural development) and have the "dis-ease" of breathing which increases their sympathetic tone. These children do not go into REM sleep and as a result their behaviors make life for everyone around them difficult because they are sleep deprived and do not know how to manage it or modulate themselves. This is true of neurotypical children as well as children on the spectrum (and just as true for their parents if they are sleep deprived because their children do not sleep!). Studies have been conducted worldwide to evaluate orofacial dysfunction and sleep disordered breathing in school age children, including recommendations for dental and orthodontic practitioners to evaluate this in every child they examine.[8,9]

On a molecular level, metabolic problems like poor iron intake or absorption (either they "don't eat it, don't absorb it and don't retain it") and low ferritin levels result in poor

capacity to oxygenate muscles. The association between low iron and sleep disordered breathing (SDB) in children has not been clearly elucidated. Iron deficiency is associated with pediatric sleep disturbances, in particular, restless leg syndrome (RLS) and periodic limb movement disorder (PLMD). Correction of iron deficiency has been shown to improve sleep disordered breathing in certain adult populations.[10]

Another reason for poor iron absorption can be due to an imbalance in the microbiome where pathogenic bacteria are using up all the iron in the gut for its own survival leaving none for the child to absorb resulting in the perpetuation of the symptoms caused by the pathogenic bacteria as well as the failure to increase their blood's ability to carry oxygen to the muscles.[11,12] The end result can be problems with sleep wake transition or a more common occurrence of muscle cramping, particularly noticed in the legs coined "restless leg syndrome" which manifests as subclinical shaking and a body that is in constant movement in sleep. When this is happening, the brain cannot rest and breathing is compromised.[13,14] A study published in 2007 by Dosman demonstrated that seventy-seven percent of the autistic children that participated had restless sleep at baseline, which improved significantly with iron therapy, suggesting a relationship between sleep disturbance and iron deficiency in children with autism spectrum disorder.[15]

Lack of Vitamin D, lack of magnesium and other essential minerals all play a role in changing the chemistry of our bodies which is reflected in how well does our body function daily. Children on the autistic spectrum often suffer from aversion to foods due to taste, texture, or color. This leads to nutrient deficiencies that need to be detected and addressed.[16]

Although not necessarily coming from an airway perspective, the book, *WHY WE SLEEP*, authored by Matthew Walker, MD, includes statistics on how sleep affects our performance. Walker defines our youthful society as more of an under-slept generation – early school, blue light and lots of screen time, not enough sleep resulting in mood changes, differences in perception, muscle tension and causing restricted range of motion. Children often hyperventilate when stressed (shallow breathing and over breathing). Clinically the patients are often breathing too much, or they are breathing incorrectly.[17] Patrick McKuen – foremost teacher of Boteyko breathing teaches we are breathing too fast and too often.[18] In his famous book *BREATH*, James Nestor writes in an easy-to-understand manner about how breathing incorrectly changes our life and technics how to correct our breathing and improve our overall health.[19]

Another book written in the 50's-60's by Dr. Alfred Fonder titled *The Dental Physician*, contributes some interesting observations on the interrelationship of the very sensitive temporomandibular joint and the autonomic nervous system (another "player" in the sleep/wake cycle as well as ability to adapt or cave under stress).[20] Fonder was a dentist working and teaching at the university of Chicago and was 50 years ahead of his time. Only now are we beginning to appreciate his brilliant, far reaching jaw concepts and his teaching about how jaw posture affects head and neck posture and spinal posture. He proposed that the mouth is essentially a powerful nervous system that is totally underappreciated by dentists, medical doctors, osteopaths and chiropractors alike. If someone had headaches, they might have been better off having utilized Fonder's approach of jaw balancing than pursuing a pharmaceutical program once any serious pathology was ruled out. Fonder wrote extensively about "dental distress syndrome" hypothesizing about the mouth and TMJ's affect on the entire body.[20]

The Central Nervous System (CNS) has allocated a certain amount of resources to deal with sensory information being received whether that be pain or a physical stressor (for example reduced oxygen intake as a result of airway compromise). It has to pay attention to that particular situation (reduced oxygen) and goes into survival mode, and as a result, other critical physiologic functions suffer. For example, poor development of the prefrontal cortex, therefore executive function, a healthy microbiome and gastrointestinal function or the ability to learn attend to instructions are only a few of the problems children in our culture, as well as children who have been diagnosed on the spectrum suffer from that might be related to airway dysfunction.[21]

In his book, Fonder documented for example, how someone's health improved when they realigned the jaw's temporomandibular joints (TMJ). He discusses how we all have adaptive capacity with room for error. The TMJ can be slightly off but if "too off" problems as simple as neck tension to as severe as psychosis may occur. Fonder conducted an experiment with women who complained of head and neck pain/TMJ pain. He recorded their symptoms and then began to change their bite demonstrating a reduction in symptomatology, including surprisingly, psychosocial issues and genitourinary problems (again, issues often shared by the autistic child and adult).[20]

Fonder relates another case report of a male child with a diagnosis of Down syndrome, a genetic diagnosis of a developmentally delayed child (13-year-old at the level of a 5-year-old); they utilized dental devices (braces, expanders) and they readily expanded his palate to see what would change. With the resultant craniofacial development that occurred over a matter of several months of wearing the appliances, the 13 year's behavior matured to an appropriate level. Their hypothesis was that he improved so much because

the pituitary gland was able to function more appropriately because of the shift in the cranial bones with the expansion of the palate. They did not consider improvement in respiratory function which would contribute to appropriate oxygenation of the prefrontal cortex encouraging neurologic maturation. Then, for an unknown reason, they abandoned the treatment, and everything regressed. As the expansion receded and the cranial bone positions reverted, his behavior regressed to that of a 5-year-old.[20] Since then, other papers have been authored about sleep-disordered breathing in the pediatric setting in a variety of genetic disorders as well as elucidating the genetic markers for the potential for sleep disordered breathing in both neurotypical and neuroatypical children.[22-24]

During sleep, neuromuscular tone supports a patent airway for breathing. A decrease in tone, or mechanical obstruction from adenoid and tonsil hypertrophy, craniofascial underdevelopment or segmental dysfunction or soft tissue imbalance can result in a tendency for the upper airway to collapse resulting in reduced air flow (which can cause oxygen desaturation) and arousal (bruxing, grinding, night terrors, waking to feed) resulting in fragmented or lack of restorative sleep which can result in failure to thrive, behavioral problems, inattention, aggression, restlessness, depression and cardiovascular problems.[25]

Inverventional orthodontics will often be recommended to promote proper bone growth and support a airway. Back in Fonder's day, dentists were not yet using more functional appliances that are available today, nor did they use "oral orthotics" to build up the bite. Rather, they used graded fillings (mercury fillings, no less) to build up the bite. Nowadays, the dentist or orthodontist will take an x-ray, assess the architecture and the mechanics of the TMJ and project where the jaw joint should be and correct the position of the lower jaw with a dental appliance. Today's dental appliances range from both traditional or newer functional metal devices to those made of a variety of plastics (or combinations thereof). The patient is then re-x-rayed to confirm the corrected position of the TMJ. This alignment correction's goal is to reduce the firing of the CNS and the patient should be relieved of symptoms of high sympathetic tone (migraine, tension headaches, neck pain, sleep disruption, behavioral aberrations).[8,26,27,28]

Dentists can play a significant role in the early detection of some craniofacial causes for sleep disordered breathing. Including the dentist as a member of the multidisciplinary team in the early evaluation of children for any of the wide variety of causes of these airway issues that might disrupt sleep should not be underestimated.[8,9,28,29]

References:

1. American Academy of Physiological Medicine and Dentistry. *Finding Connor Deegan*. Suffern, NY 2014 April: Oy https://www.youtube.com/watch?v=Sk5qsmRyVcE&t=45s.

2. Sinha D, Guilleminault C. Sleep disordered breathing in children. *Indian J Med Res*. 2010 Feb;131:311-20. PMID: 20308756. links to full text: https://pubmed.ncbi.nlm.nih.gov/20308756/.

3. Marcus CL. Sleep-disordered breathing in children. *Am J Respir Crit Care Med*. 2001 Jul 1;164(1):16-30. doi: 10.1164/ajrccm.164.1.2008171. PMID: 11435234.

4. Hotwani K, Sharma K, Jaiswal A. Evaluation of tongue/mandible volume ratio in children with obstructive sleep apnea. *Dental Press J Orthod*. 2018 Aug 1;23(4):72-78. doi: 10.1590/2177-6709.23.4.072-078.oar. PMID: 30304156; PMCID: PMC6150701. https://www.ncbi.nlm.nih.gov/pmc/articles/PMC6150701/.

5. Chu B, Marwaha K, Sanvictores T, et al. Physiology, Stress Reaction. [Updated 2021 Sep 18]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2022 Jan-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK541120/.

6. Boyd K, Saccomanno S, Lewis CJ, Coceani Paskay L, Quinzi V, Marzo G. Myofunctional therapy. Part 1: Culture, industrialisation and the shrinking human face. *Eur J Paediatr Dent*. 2021;22(1):80-81. doi: 10.23804/ejpd.2021.22.01.15. PMID: 33719489.

7. Malkani M. *Simple & Safe Baby-Led Weaning*. Rockbridge Press; Emeryville,CA 2020.

8. Al Ehaideb AA, Almufadhi NM, Ab Alhassn GM, Fallatah AA, Adnan S, Alsubaie AA. Sleep-disordered breathing among Saudi children seeking orthodontic treatment. *J Family Med Prim Care*. 2021 Jan;10(1):205-212. doi: 10.4103/jfmpc.jfmpc_1918_20. Epub 2021 Jan 19. PMID: 34017727; PMCID: PMC8132852.

9. Abdalla M, Halabi M, Kowash M, Hussein I, Khamis A, Salami A. Sleep-disordered breathing in children and adolescents seeking paediatric dental care in Dubai, UAE. *Eur Arch Paediatr Dent*. 2022 Jun;23(3):485-494. doi: 10.1007/s40368-022-00697-8. Epub 2022 Feb 27. Erratum in: *Eur Arch Paediatr Dent*. 2022 Mar 21;: PMID: 35220544.

10. Kerstein R, Stimpson P, Caulfield H, Ellis G. Iron deficiency and sleep disordered breathing in children–cause or effect? *Int J Pediatr Otorhinolaryngol*. 2009 Feb;73(2):275-80. doi: 10.1016/j.ijporl.2008.10.023. Epub 2008 Dec 10. PMID: 19081645.

11. Seyoum Y, Baye K, Humblot C. Iron homeostasis in host and gut bacteria - a complex interrelationship. *Gut Microbes*. 2021 Jan-Dec;13(1):1-19. doi: 10.1080/19490976.2021.1874855. PMID: 33541211; PMCID: PMC7872071.

12. Pickard JM, Zeng MY, Caruso R, Núñez G. Gut microbiota: Role in pathogen colonization, immune responses, and inflammatory disease. *Immunol Rev.* 2017 Sept; 279(1):70-89. doi: 10.1111/imr.12567. PMID: 28856738; PMCID: PMC5657496.

13. Tilma J, Tilma K, Norregaard O, Ostergaard JR. Early childhood-onset restless legs syndrome: symptoms and effect of oral iron treatment. *Acta Paediatr*. 2013 May;102(5):e221-6. doi: 10.1111/apa.12173. Epub 2013 Mar 6. PMID: 23360128.

14. Rosen GM, Morrissette S, Larson A, Stading P, Barnes TL. Does improvement of low serum ferritin improve symptoms of restless legs syndrome in a cohort of pediatric patients?. *J Clin Sleep Med*. 2019;15(8):1149—1154.

15. Dosman CF, Brian JA, Drmic IE, et al. Children with autism: effect of iron supplementation on sleep and ferritin. *Pediatr Neurol*. 2007;36(3):152—158.

16. Cermak SA, Curtin C, Bandini LG. Food selectivity and sensory sensitivity in children with autism spectrum disorders. *J Am Diet Assoc*. 2010 Feb;110(2):238-46. doi: 10.1016/j.jada.2009.10.032. PMID: 20102851; PMCID: PMC3601920.

17. Walker M. *Why We Sleep*. Scribner Books; 2017. ISBN 978-0-241-26906-0.

18. McKuen P. *The Breathing Cure: Develop New Habits for a Healthier, Happier, and Longer Life*. Humanix Books; 2021.

19. Nestor J. *Breath: The New Science of a Lost Art*. Riverhead Books; 2020.

20. Fonder AC. *The Dental Physician* [Second 2nd Revised Edition]. Medical-Dental Arts; 1985.

21. Mir E, Kumar R, Suri TM, Suri JC, Venkatachalam VP, Sen MK, Chakrabarti S. Neurocognitive and behavioral abnormalities in Indian children with sleep-disordered breathing before and after adenotonsillectomy. *Lung India*. 2019 Jul-Aug;36(4):304-312. doi: 10.4103/lungindia.lungindia_398_18. PMID: 31290415; PMCID: PMC6625250.

22. Zaffanello M, Antoniazzi F, Tenero L, Nosetti L, Piazza M, Piacentini G. Sleep-disordered breathing in paediatric setting: existing and upcoming of the genetic disorders. *Ann Transl Med*. 2018 Sep;6(17):343. doi: 10.21037/atm.2018.07.13. PMID: 30306082; PMCID: PMC6174189.

23. Lu A, Luo F, Sun C, Zhang X, Wang L, Lu W. Sleep-disordered breathing and genetic findings in children with Prader-Willi syndrome in China. *Ann Transl Med*. 2020 Aug;8(16):989. doi: 10.21037/atm-20-4475. PMID: 32953789; PMCID: PMC7475489.

24. Mainieri G, Montini A, Nicotera A, Di Rosa G, Provini F, Loddo G. The Genetics of Sleep Disorders in Children: A Narrative Review. *Brain Sci*. 2021 Sep 23;11(10):1259. doi: 10.3390/brainsci11101259. PMID: 34679324; PMCID: PMC8534132.

25. Kanthur N, Halbower AC. Sleep disordered breathing in children. In: Bajaj L, Hambidge SJ, Kerby G, Nyquist AC, eds. *Berman's Pediatric Decision Making* (Fifth Edition). Mosby; 2011:760-762.

26. Farsi NM. Symptoms and signs of temporomandibular disorders and oral parafunctions among Saudi children. *J Oral Rehabil*. 2003 Dec;30(12):1200-8. doi: 10.1111/j.1365-2842.2003.01187.x. PMID: 14641664.

27. Vierola A, Suominen AL, Eloranta AM, Lintu N, Ikävalko T, Närhi M, Lakka TA. Determinants for craniofacial pains in children 6-8 years of age: the PANIC study. *Acta Odontol Scand*. 2017 Aug;75(6):453-460. doi: 10.1080/00016357.2017.1339908. Epub 2017 Jun 16. PMID: 28622039.

28. Leibovitz S,Haviv Y, Sharav Y, Almoznino G, Aframian D, Zilberman U. Pediatric sleep-disordered breathing: Role of the dentist. *Quintessence Int*. 2017;48(8):639-645. doi: 10.3290/j.qi.a38554. PMID: 28681043.

29. Heit T, Tablizo BJ, Salud M, Mo F, Kang M, Tablizo MA, Witmans M. Craniofacial Sleep Medicine: The Important Role of Dental Providers in Detecting and Treating Sleep Disordered Breathing in Children. *Children* (Basel). 2022 Jul 15;9(7):1057. doi: 10.3390/children9071057. PMID: 35884041; PMCID: PMC9323037.

The chiropractor's role in the treatment of sleep disordered breathing

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ABSTRACT

As sleep problems are a common issue for infants, all professionals work with these children with short- and long-term benefits in mind. The purpose of this clinically oriented article is to acknowledge, investigate and provide a professional commentary on some of the key parameters that should be a priority for chiropractors working with children with sleep disorders.

Key terms: Disordered sleep, chiropractic, infants, children, respiratory cycle.

Introduction

Childhood sleep disordered breathing has become an epidemic problem throughout the world, with 95% of the children with obstructive sleep apnea failing to be diagnosed. "700,000 Aussie kids under 10 have sleep problems which goes up to 1.9 million in the UK, 11 million in the USA and half a billion in Asia."[1] As the quality of children's sleep deteriorates, the rates of childhood obesity, anxiety and behavioral disorders have been on the rise.

Which healthcare specialty is best suited to diagnose and treat childhood sleep disorders? An infant's first healthcare providers may be a midwife, doula, lactation consultant and/or a pediatrician. As they get older, they may be referred to an ear, nose and throat (ENT) specialist, pediatric dentist, speech language pathologist, occupational therapist, oral myofunctional therapist, or a behavior therapist. Another healthcare provider whose care spans children of all ages and has the appropriate training to identify this problem is the pediatric chiropractor. The pediatric chiropractor is in a unique position to not only identify the problem, but to make the appropriate referrals while rendering care to the child in a collaborative relationship to resolve the condition. According to a study by Moore, et al., that although snoring and sleep apnea may be the most common indication of a respiratory sleep disorder in a child, one quarter of children presenting to a sleep clinic for evaluation will have a second sleep diagnosis, which is often non-respiratory in nature. They recommend that clinicians working in this arena must be prepared to recognize, evaluate, and manage sleep disorders across the lifespan of the patient.[2]

Defining the Issue

Sleep disordered breathing is defined as a blockage of all or part of the airway. There is a spectrum of sleep disorders, ranging from open mouth breathing to upper airway resistance syndrome, to snoring and to obstructive sleep apnea. When open mouth breathing is observed in a newborn baby, it is often accompanied by a recessed mandible and possible tongue, lip and/or buccal ties. Snoring can be heard and may be indicative of obstruction. It is the author's opinion that snoring should not be considered normal in an infant (or at any age). Upper airway resistance will sometimes present as noisy breathing. Other types of sleep disordered breathing are respiratory effort-related arousals (RERAs) and parasomnias. RERAs are defined as changes in upper airway pressure which limit the flow of air during each breath in the later sleep stages and rapid eye movement (REM) sleep. Parasomnias include sleepwalking, night terrors, unusual movements, teeth grinding, nightmares and sleep-related eating disorders.

There are many signs and symptoms of sleep disordered breathing, starting with infancy, moving through childhood and into adulthood. Many of these symptoms are either overlooked, or the symptoms are treated but the cause is never addressed. At birth, an inability to latch or breastfeed successfully (meaning transfer sufficient milk to sustain themselves) can be a red flag to assess an infant for sleep disordered breathing. One cause of either or both aforementioned issues could be tethered oral tissue syndrome, aka "tongue-tie."[3,4] Cranial distortions can also cause latching issues.[5] A recessed mandible can be observed in either one of these conditions. The child may also have a heightened gag reflex which would be an intact neurologic mechanism to prevent aspiration of liquids when the integrity of the suck, swallow, breath synchrony is impaired.[6] As the child grows, an open mouth posture and venous pooling under the eyes may be observed, as well as an architecturally narrow face or poor midface development.[7]

Parental description

The parents may report that the child has many bedtime antics to avoid going to sleep. Once asleep, the child may be very restless, awaken through the night, have enuresis,

nightmares, or night terrors. Sometimes they will sleep for long periods of time but never seem well rested.[8] Speech issues with or without tongue thrust (pressing the tongue up against the teeth or between them while swallowing) are very common. These children are often picky eaters and prefer to consume juice or some other type of sugary drink to "keep them going" since they are exhausted. Behaviors can be very challenging with these children.[9]

Sleep disordered breathing in infants can be difficult to diagnose due to the varied signs and symptoms. These babies often do not like to lie on their backs nor their stomachs, preferring to be held. The caregiver will often report that the baby will only lie on their stomach if they are on someone's lap, thus not in a completely flat position. When nursing, they will often pull on or pull off the nipple, fatigue quickly and either fuss or fall asleep, exhausted from their efforts, before transferring an adequate amount of milk to satiate themselves. They may have a narrow gape and/or shallow latch and dribbling while nursing is also common. The mother will often report that she must supplement nursing, and the provider needs to help her discern whether her milk supply is insufficient or whether the infant is unable to transfer milk therefore failing to stimulate her milk supply. The mother may also report that breastfeeding is painful and that the infant cannot open their mouth wide enough to get a deep, secure latch. This baby will often have a difficult time taking a pacifier as they cannot hold onto it, due to a tongue thrust and/or shallow suck.

Sleep disordered breathing can also present itself as noisy breathing or a light snore–a cause for alarm in an infant. Infants with sleep disordered breathing are also often diagnosed with gastroesophageal reflux (GER).[10] These conditions–inadequate milk transfer, a tongue thrust, a shallow latch or weak suckle, and gastroesophageal reflux disease are often accompanied by aerophagia. This is when the baby is taking in air while feeding, whether it be by breast or bottle, which can be correlated with sleep disordered breathing.[11]

Long-term problems

If sleep disordered breathing is not diagnosed and treated in infancy, it may progress to more serious problems as a toddler, school age child or adolescent. This child will often be observed with open mouth breathing while awake or sleeping. This child may wake frequently through the night. One example would be a toddler who is waking frequently (8-10x at night) to breast or bottle feed. While sleeping, these children may have nightmares, night terrors, restlessness, sleepwalking and persistent enuresis (despite demonstrating bladder control while awake). This child can be very hard to put to bed because they are in a constantly elevated sympathetic state ("fight or flight"). While eating, they will often refuse anything other than soft processed foods such as macaroni and cheese, crackers, or processed chicken nuggets. They are often a slow eater, have an aversion to chunky or chewy textures, and may have a heightened gag reflex (in some cases, causing them to vomit their food). They will constantly crave simple carbohydrates which will perpetuate the sympathetic state. They will often have a nasal voice because of swollen adenoids and/or tonsils. They will be prone to colds and allergies, venous pooling under the eyes and a narrow chin. Behavior issues may start to emerge: they will often be emotional and predisposed to outbursts or anger. When they are of school age, they will frequently be diagnosed with attention deficit hyperactivity disorder (ADHD), attention deficit disorder (ADD) or oppositional defiant disorder (ODD), with poor focus, inability to concentrate and distractibility as primary symptoms complicated by defiant and impulsive behaviors.[12] Speech can continue to be an issue.[13]

Respiration cycle

The suck, swallow, breath synchrony evolves in utero, with swallowing beginning at 11 weeks. The organized suck/swallow pattern emerges by 32 weeks in utero. The tongue raised at rest and resting on the palate creates the shape of the palate. The palate is the bottom of the maxillary sinuses and the shape of the palate helps determine the size of the airway. Cranial-sacral therapy as taught by sacro-occipital technique (SOT) provides training on evaluation and treatment.[14] Lips closed and a closed mouth posture function as future braces for the teeth, allowing them to come in naturally. A correct suck/swallow position is lips closed with tongue resting on palate while nasal breathing.[15]

All twelve cranial nerves are involved with breastfeeding, but there are seven of the twelve which are critical for successful breastfeeding. As a baby turns their head towards the nipple, they engage accessory cranial nerve (XI). Facial nerve (VII) and trigeminal nerve (V) are used to open the mouth to latch onto the nipple. Hypoglossal nerve (XII) is needed to push the tongue up on the nipple to stimulate milk production. Finally, the milk needs to be delivered to the back of the throat to swallow and the nerves utilized for this are the glossopharyngeal nerve (IX), vagus nerve (X) and trigeminal nerve (V). Craniocervical dysfunctions can impair the correct processing of the cranial nerves.[16] SOT practitioners with cranial training can be effective for helping to resolve cranial nerve issues. The correct pattern for nursing is suck, swallow, breathe, suck swallow, breathe, over and over.[17]

Bottle feeding can cause many issues that may not show up until the baby is a bit older. When a baby bottle feeds, the milk flows into the mouth more easily. The baby does not need to open their mouth as wide as they would

breastfeeding. The tongue will often thrust forward to control the flow of milk. All of these patterns may lead to cranial-facial developmental changes that they could carry for the rest of their life. Since the tongue does not need to push up on the palate, the same as during breastfeeding, the palate may not widen out and develop as fully as that of a breastfed baby. This can cause the palate to be high and arched which, in turn, potentiates open mouth breathing, crooked teeth and a need for braces. They could also develop a hooked nose, narrow chin and a smaller airway, causing them to be more prone to ear infections, allergies and asthma.[18]

Associated issues

One reason why breastfeeding can be difficult if not impossible is Tethered Oral Tissue Syndrome, aka TOTS or ankyloglossia. TOTS can be an anterior tie, posterior tie, lip or buccal tie. The definition of a tongue tie is restricted mobility as a result of a short lingual frenum, a condition often affecting breastfeeding, but not always. Evaluating for a tongue tie can be easily done during an examination. The doctor places the baby in a supine position. While wearing gloves, the doctor places two fingers under each side of the tongue and lifts it up towards the palate while an assistant (or parent) pulls the chin down. The tongue should be able to lift up to the palate. This is when the doctor will observe the frenulum, checking that it does not pull up the floor of the mouth or blanch too much. Not all frenum need be revised with a frenectomy. It is the author's opinion that performing cranial-sacral work on a baby will often help the baby to nurse, but a minimal to mildly restrictive frenum does not always need a revision, nor would it classify as "ankyloglossia." If a baby ultimately needs a frenectomy, manual therapy ("body work" as it is referred to colloquially) with soft tissue therapies, chiropractic adjusting and cranial-sacral work may be helpful to ready the baby for the procedure to optimize the outcome.[19] Keeping up with manual therapy after the procedure is very important to help the baby integrate the changes that have been made neurologically and reduce their compensatory motor patterns and to further reduce any dural tension as a result of the tethered oral tissues.[20] If this condition is not corrected (with or without surgery), there is a possible cascade of symptomatology that can occur. The infant or child may display open mouth breathing, develop a narrow palate/face, frequent illnesses that can lead to snoring and eventually to obstructive sleep apnea as the tongue slides back in the airway rather than remaining up on the palate.

Nasal breathing is what we are designed to do — but what are the effects of nasal versus mouth breathing? Very simply, nasal breathing warms and humidifies air, filters allergens and microbes, creates nitric oxide, increases our ability to absorb oxygen, regulates blood pressure and keeps us in a calm parasympathetic state. Mouth breathing, on the other hand, results in dry mouth, bad breath, snoring, fatigue, brain fog, dental caries and continues to elicit a sympathetic response. Craniofacial development is also affected. A nasal breather tends to develop a wide face, good cheekbones, alert eyes and a straight nose. A mouth breather is prone to develop a narrow face, crooked teeth, crooked nose, head forward posture, tired eyes, droopy eyes, and venous pooling under the eyes.[21]

Sleep stages

It is important that the chiropractor understand all the stages of sleep and the hormonal implications of getting proper or restorative sleep, versus sleep interrupted by sleep disordered breathing. Sleep is usually divided into non-rapid eye movement (NREM) and rapid eye movement (REM). Adults cycle through four to five times a night with each stage lasting 90-120 minutes. Babies and children cycle through more often with their REM sleep lasting longer depending on the number of hours that they sleep.[22]

NREM has four stages. Stage one involves falling asleep during which the heart rate will slow down but the person is still easily aroused by light and sound. Stage two is the longest phase of sleep. At this point, the muscles will relax and snoring may start. Our brains are at work consolidating all the learning from the day. Stages three and four are the deepest phases of NREM sleep. During these stages, temperature, heart rate and breath rate all decrease, and parasomnias and enuresis can occur. Long-term memory is consolidated, and tissue repair and release of growth hormone take place.[22]

During REM, procedural and spatial memory are created (often referred to as the "dream stage"). Physiologically, the body is paralyzed during this stage, and rate of breath, heart rate and blood pressure all increase, and toxins are removed from the brain while brain activity increases.[22]

Time spent in REM vs NREM sleep changes as a child grows. While a six-month-old baby spends their sleeping time equally split between REM and NREM, a five-year-old will only spend 30% of their time in REM and 70% in NREM sleep. By the later teenage years, only 20% of sleep is spent in REM and 80% in NREM which continues throughout adulthood. One reason for this is that neuronal pathways are laid down during REM sleep and synaptic pruning occurs during NREM sleep. Development of the brain starts in the back of the brain, the primal brain, and moves into the front cerebral cortex as maturity takes place. This is why getting proper sleep is important for critical thinking to develop.[23]

Hormonal activity during these stages is deeply affected by sleep disordered breathing. Secretion of antidiuretic hormone (ADH), atrial natriuretic factor, leptin, ghrelin,

somatotropin, melatonin, and cortisol are all affected. During normal sleep, ADH is released which inhibits urination while in deep sleep. Atrial natriuretic factor, a hormone secreted by the heart to regulate salt-water balance and blood pressure, is inhibited by sleep apnea which in turn inhibits the release of ADH.[24] Also, secretion of leptin, which inhibits hunger, is decreased with sleep disordered breathing, and ghrelin, a hormone stimulating the drive to eat, is increased. Somatotropin (growth hormone) and melatonin are both decreased, while cortisol is increased, with sleep disordered breathing.[25]

Irregularities in breathing during sleep can cause permanent damage to health, including brain damage with an up to 10-point loss in intelligence quotient. Neurocognitive deficits include impaired attention, focus, reasoning, and problem solving. Prolonged sleep issues reduce gray matter in the brain, and low blood oxygen impairs the immune system but also the growth of a child.[26,27]

There is also a strong link between inadequate sleep during childhood, and an increased risk of Alzheimer's disease in adulthood. The glymphatic system in our brain is the lymphatic system for the glial cells, most active during the deepest phases of NREM sleep. The glial cells shrink by 60 per cent during these phases to accommodate space around neurons to allow cerebral spinal fluid to flow more easily and flush out metabolites from the day's neuronal activity. This process during NREM sleep also cleans out amyloid proteins linked to Alzheimer's disease.[28]

Other key parameters: Several environmental factors can affect the quality of a child's sleep. It is best to provide a dark room for maximum production of melatonin. Screen time prior to sleep should be kept to a minimum, and the use of blue light glasses can improve sleep quality and duration. Children's sleep is affected by artificial sweeteners and food dyes, and these chemicals should be eliminated and replaced by whole foods and a minimum of sugars.

Pacifiers should be discouraged by six months of age. Prolonged use of a pacifier can affect formation of the jaw and contribute to open mouth breathing. A good alternative is the Myo Munchee, a medical grade silicone device invented by a dentist in the 1960's and carried on by his daughter, Mary Bourke, chiropractor. It helps train proper suck/swallow, stops thumb sucking, promotes nasal breathing, supports cranial-facial growth and healthy oral hygiene. It can be used with babies as young as six months (Bebe Munchee) and comes in many sizes to accommodate the growing child.[29]

Conclusion

Chiropractors may play a key role in the field of sleep disordered breathing and are often a part of a team to help with nursing issues such as latching, constipation, colic or GERD. The collaborative network often includes neuro-muscular dentists, orthodontists, pediatric dentists, oromyofunctional therapists, speech and language pathologists, occupational therapists, ENT's, behavior specialists, lactation consultants, midwives, doulas, and naturopaths. A chiropractic exam complements most pediatric exams, evaluating for infant reflexes, evaluation of the suck and cranial-sacral assessment to determine the presence of tension in the dural sheath. Chiropractors can also screen for tongue, lip, and buccal ties and can perform pre and post frenectomy work if needed. The treatment may include chiropractic adjusting, cranial sacral work, teaching use of Myo Munchees and educating families about environmental and nutritional correlations for quality sleep. This team approach is often the optimal way forward to assist children in fully functional feeding and sleeping mechanics.

The question always to be explored is, "Where is your tongue?" The goal should be lips sealed, with the tongue on the palate, and nasal breathing.

References:

1. Moore S. Sleep Wrecked Kids. New York: Morgan James Publishing;2020.

2. Moore M, Allison D, Rosen CL. A review of pediatric non-respiratory sleep disorder. *Chest* Oct 2007;130(4):1252-1262.

3. Alan E. Frenotomy may help resolve breastfeeding problems due to tongue-tie. *The Journal of Pediatrics* May 2015;166(5):1320-1323.

4. Tobey AH, Kozar A. Frequency of somatic dysfunction in infants with tongue-tie: a retrospective chart review. *The AAO Journal* 2018;28(4):10-14.

5. Westcott N. The use of cranial osteopathy in the treatment of infants with breastfeeding problems or sucking dysfunction. *The Australian Journal of Holistic Nursing* 2004;11(1):25-32.

6. Coryllos E, Genna CW, Salloum AC. Congenital tongue-tie and its impact on breastfeeding. *American Academy of Pediatrics*, Section on Breastfeeding 2004:1-6.

7. Pacheco MCT, Fiorott BS, Finck NS, Martins De Araujo MT. Craniofacial changes and symptoms of sleep-disordered breathing in healthy children. *Dental Press J Orthodontics* May-June 2015;20(3)80-87.

8. Chervin RD, Hedger K, Dillon JE, Pituch KJ. Pediatric sleep questionnaire (PSQ):validity and reliability of scales for sleep-disordered breathing, snoring, sleepiness, and behavioral problems. *Sleep Medicine* Feb 2000; Vol 1(1):21-32.

9. Bonuck K, Freeman K, Chervin RD, Xu L. Sleep disordered breathing in a population-based cohort: behavioral outcomes at 4 and 7 years. *Pediatrics*. 2012;129(4):e857-e865.

10. Sinha D, Guilleminault C. Sleep disordered breathing in children. *Indian J Med Res* Feb 2010; Vol 131:311-320.

11. Machedo R, Woodley FW, Skaggs B, DiLorenzo C, Splaingard M, Mousa H. Gastroesophageal reflux causing sleep interruptions in infants. *J Pediatr Gastroenterol Nutr* 2013;56(4):431-5.

12. Rosen CL, Storfer-Isser A, Taylor HG, Kirchner HL, Emancipator JL, Redline S. Increased behavioral morbidity in school-aged children with sleep-disordered breathing. *Pediatrics* Dec 2004;114(6):1640-1648.

13. Mohammed D, Park V, Bogaardt H, Docking K. The impact of obstructive sleep apnea on speech and oral language development: a systematic review. *Sleep Medicine* May 2021;81:144-153.

14. Blum CL.Chiropractic and dentistry in the 21st century. *Cranio* 2004; 22(1):1-3.

15. Campanha SMA, Martinelli R, de Castro L, Palhares DB. Position of lips and tongue in rest in newborns with and without ankyloglossia. *CoDAS* 2021;33(6):1-5.

16. Vallone S. Evaluation and Treatment of Breastfeeding Difficulties Associated with Cervicocranial Dysfunction: A Chiropractic Perspective. *JCCP* 2016 (Dec); 15(3): 1301 -1305.

17. Sakalidis VS, Geddes DT. Suck-swallow-breathe dynamics in breastfed infants. *J Human Lact* May 2016;32(2): 201-211.

18. Radzi Z, Yahya NA. Relationship between breast-feeding and bottle feeding to craniofacial and dental development. *Annals of Dentistry* University of Malaya 2005;12(1):9-17.

19. Ghaheri BA, Cole M, Fausel SC, Mace JC. Breastfeeding improvement following tongue-tie and lip-tie release: a prospective cohort study. *Laryngoscope* May 2017;127(5):1217-1223.

20. Vallone S. Chiropractic evaluation and treatment of musculoskeletal dysfunction in infants demonstrating difficulty breastfeeding. *JCCP* 2004; 6(1):349-368.

21. Sarawata S, Mali L, Sinha A, Nanda SB. Effect of naso-respiratory obstruction with mouth breathing on dentofacial and craniofacial development. *Orthodontic Journal of Nepal* 2018;8(1):22-27.

22. Boyce R, Williams S, Adamantidis A. REM sleep and memory. *Curr Opin Neurobiol* June 2016;44:167-177.

23. Esposito M, Antinolfi L, Gallai B, Parisi L, Roccella M, Marotta R, Lavano SM, Mazzotta G, Precenzano F, Carotenuto M. Executive dysfunction in children affected by obstructive sleep apnea: an observational study. *Neuropsychiatric Disease and Treatment* 2013;9:1087-1094.

24. Capdevila OS, McLaughlin Crabtree V, Kheirandish-Gozal L, Gozal D. Increased morning brain natriuretic peptide levels in children with nocturnal enuresis and sleep-disordered breathing: a community-based study. *Pediatrics* 2008;121(5):e1208-e1214.

25. Saaresranta T, Polo O. Sleep disordered breathing and hormones. *European Respiratory Journal* July 2003;22:161-172.25.

26. Kaemingk KL, Pasvogel AE, Goodwin JL, Mulvaney SA, Martinez F, Enright PL, Rosen G, Morgan WJ, Fregosi RF, Quan SF. Learning in children and sleep disordered breathing: findings of the Tucson children's assessment of sleep apnea (TuCASA) prospective cohort study. *Journal of the International Neuropsychological Society* Feb 2004;9(7):1016-1026.

27. Spooner R, Lushington K, Keage HAD, Blunden S, Kennedy JD, Schembri M, Wabnitz D, Martin JA, Kohler MJ. Cognition, temperament, and cerebral blood flow velocity in toddlers and preschool children with sleep-disordered breathing or behavioral insomnia of childhood. *Sleep Med* May 2016; 21:77-85.

28. Ju Yo-El S, Finn M, Sutphen CL, Herries EM, Jerome GM, Ladenson JH, Crimmons DL, Fagen AM, Holtzman DM. Obstructive sleep apnea decreases central nervous system-derived proteins in the cerebrospinal fluid. *Ann Neurol* July 2016;80(1):154-9.

29. Bourke M, Cole C. Why they suck. The Junction: *Munchee*.

Lumbopelvic Presentations in Pregnancy Through the Lens of Sacro Occipital Technique

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ABSTRACT

Chiropractic care is commonly sought by women during pregnancy. While many motivating factors exist for seeking care, a common one is the desire to have a pregnancy that is comfortable and allows for life's activities without pain and limitations. Of added concern is the desire to have care that is both gentle and efficacious. As such, practitioners must remain vigilant with the rapid and natural biomechanical changes that occur in the lumbopelvic and sacroiliac regions during pregnancy. This due diligence must include the awareness of the relevant neuromusculoskeletal structures as well as the chiropractic technique approaches that best suit the individual pregnant patient. Sacro Occipital Technique (SOT®) is a low force chiropractic technique that addresses the unique biomechanical sequelae of the pregnant pelvis. Presentations like lumbar facet syndrome with iliopsoas hypertonicity, gluteus medius instability, and piriformis syndrome are common concomitant presentations that manifest during pregnancy. These clinical presentations not only warrant the need for chiropractic care, but also, the consideration of SOT® as a methodology that is well positioned to palliate these specific concerns. The goal of this article is to present SOT in its foundational context and provide clinical relevance for its specific use during pregnancy.

Key words: Sacro Occipital Technique, SOT, pregnancy, instability, pelvic girdle dysfunction, low back pain, facet syndrome, piriformis syndrome, iliopsoas, gluteus medius, Category II.

Introduction

The art and science behind the principles and practice of Sacro Occipital Technique (SOT®) are the life's work of Major Bertrand DeJarnette. Born in Greenridge, MO in 1899, Dr. DeJarnette spent the bulk of his lifetime in Nebraska City, NE where the confluence of his education and clinical practice brought SOT® to the chiropractic profession.[1] With a penchant for mechanical engineering, Dr. DeJarnette worked in the automotive industry in Detroit, MI. Occupational injury led to his need to rehabilitate and heal and to a fascination with the intricacies of human structure and function. Upon completing his D.O. degree in 1922 from Dearborn College of Osteopathy in Elgin, IL, Dr. DeJarnette also completed his D.C. education from Nebraska Chiropractic College.[1]

In his lifetime of practice, Dr. DeJarnette made clinical observations in the areas of vasomotor changes, visceral pain patterns, postural distortions, and pain localization. It is the elaboration and organization of these observations that have created the foundation of the functional principles of SOT®.

SOT® Methods and Patterns

With an original twenty-one diverse categories of presentation, Dr. DeJarnette effectively distilled patterns of clinical presentation into three main clinical categories. These categories were threaded together with the physiologic interplay of the sacro-occipital relationship, flow of cerebrospinal fluid, and the load-bearing adaptability of the sacroiliac joint. The critical component of this physiologic thread is the connectedness of the primary respiratory mechanism (inhalation/exhalation cycle) with the secondary respiratory mechanism. The secondary respiratory mechanism relies upon the cranio-sacral pump as it mobilizes cerebrospinal fluid from the cranial vault to the sacrum. In the SOT® practice paradigm, corrections of all three categories address the function of these mechanisms.

Three Clinical Categories in SOT®

Each of the three categories have characteristic symptoms that provide the chiropractor with crucial clinical signs that direct care and intervention (Table 1). In pregnancy, biomechanical changes commonly cascade into a Category II type of presentation. Pursuant to these changes, patients can often present with low back pain that can be attributed to gluteus medius weakness, changes in iliopsoas tonicity, lumbar facet dysfunction, and/or piriformis syndrome.

It is imperative that the practitioner be familiar with the clinical presentation of these concomitant conditions to appropriately utilize the SOT® Category II corrections

Category I	Torsion/misalignment of the sacrum around the Y-axis Vasomotor/circulatory changes noted at specific segmental levels CSF stagnation owing to cranio-sacral pump dysfunction Palpatory hypertonicity in scalene musculature Bilateral rib head pain or fixation A-P postural sway Increased heel tension
Category II	Instability of the articulations of the pelvis Hypertonicity of the pelvic stabilizing muscles Increased global postural tone — lumbar, thoracic, and cervical Unilateral rib head pain or fixation Lateral postural sway
Category III	Due to an unresolved Category II Mechanical stress transferred to lumbar spine anatomy Facet syndrome Discogenic pathology Antalgic posture

Table 1: Overview of categories.

(Figure 1). In addition to preventing injury and pain, chiropractic care focused on stabilizing the pregnant pelvis may contribute to overall better pregnancy outcomes and support optimal fetal positioning.[2]

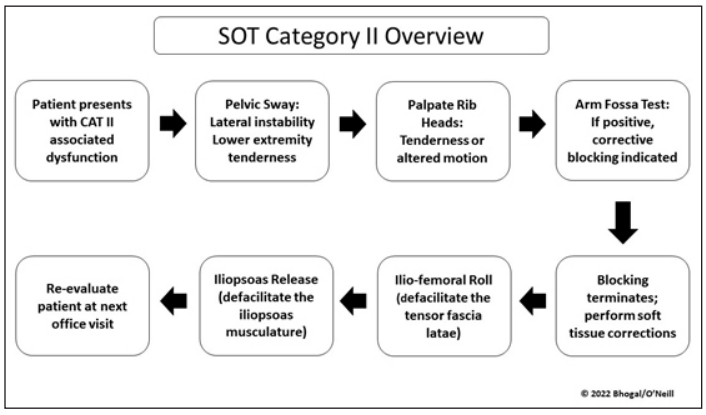

Figure 1.

Associated Lumbopelvic Presentations in Pregnancy
Low back pain (LBP) is commonly experienced during pregnancy and can interfere with a patient's quality of life.[3] LBP may present as one of two different patterns commonly described as pelvic girdle pain (PGP) and lumbar pain (LP), though some patients present with both.[4]

Many factors contribute to pregnancy-related LBP. It is commonly accepted that biomechanical factors such as weight gain, increased abdominal diameter, and the anterior shift of the body's center of gravity increase stress on the lumbar spine and pelvis. Axial loading of the intervertebral discs leads to decreased height and compression of the spine.[5] This, in conjunction with postural adaptations, including lumbar lordosis, can increase the stress placed on lumbar facet joints.[4]

Additionally, LBP has been correlated to weakness and dysfunction of core muscle groups, including changes in the abdominal wall musculature, weakness of the gluteus medius and pelvic floor dysfunction.[5,6] This muscle weakness, commonly seen in pregnancy and post-partum, alters body mechanics and increases the risk of injury as atrophied tissues are less tolerant of physical stresses.[7]

Gluteus Medius Weakness in Pregnancy
In addition to hip abduction, the gluteus medius assists with hip internal rotation and hip external rotation when the knee is in extension. Gluteus medius acts from the femur to stabilize the pelvis and maintain the trunk upright when standing on one leg, running, and walking when one leg is off the ground.[8]

Gluteus medius weakness is common in pregnancy and may present with trendelenburg gait.[6] A trendelenburg gait results from a defective hip abductor mechanism that causes drooping of the pelvis to the contralateral side while walking. This lateral shifting of forces, affectionately referred to as a "pregnancy waddle," not only stresses the sacroiliac and lumbar facet joints but also results in postural compensations throughout the body. These compensations often manifest symptomatically in the thoracolumbar, cervicothoracic, and craniocervical junctions.

Gluteus medius strain can present as low back pain either due to facet joint irritation relating to a trendelenburg gait or it can present as referred pain from the gluteus medius itself.[6] Referred pain from the gluteus medius may present with a myotogenous pain pattern in the lumbosacral region, posterior iliac crest and into the buttocks and the posterior lower extremity.

In some cases, however, despite gluteus medius weakness, there is no observable alteration in gait. Instead, ankle plantar flexors, hip abductors, and hip extensors compensate for the weak gluteus medius.[9] Additional stress on these structures often presents with clinical symptoms. Hypertonicity of the tensor fascia latae is commonly present and often reported by the patient as lateral "hip pain."

Iliopsoas Muscle in Pregnancy
What we commonly refer to as the iliopsoas muscle is made up of three separate structures: the major and minor psoas muscles and the iliacus muscle. Proper neuromechanical

function of this complex is essential for maintaining correct lumbar posture as well as stabilizing the hip and pelvis through gait.[10] The iliopsoas flexes the hip and externally rotates the femur as well as influences the lordotic curve. Contraction of the iliopsoas muscle flexes the trunk and inclines it from the contraction side.[11]

In pregnancy, as the center of gravity shifts forward and the pelvis tilts anteriorly, the iliopsoas engages to stabilize the lumbar spine and pelvis. Resultant hypertonicity of the iliopsoas muscle is common. Subluxations in the lumbar spine or pelvis may result in asymmetric hypertonicity which contribute to postural distortions and increased stress on intervertebral discs and lumbar facets.

Facet Syndrome in Pregnancy

Facet joints resist much of the intervertebral shear force and share in resisting the intervertebral compressive force, especially in lordotic postures. In rotation, the facet capsular ligaments undergo significant strain as they protect the intervertebral discs by preventing excessive movement.[12]

Lumbar facet syndrome occurs secondary to repetitive overuse and microtrauma, spinal strains, torsional forces, poor body mechanics, obesity, and intervertebral disc degeneration.[13] It is a common cause of low back pain in pregnancy due to increased weight gain, changes in posture, and altered muscle patterns, which frequently result in sub-optimal compensatory biomechanics. Clinically, lumbar facet syndrome may vary in its presentation. The patient may report localized pain and/or pseudo-radicular pain with variable referral patterns.[12] Examination may reveal decreased extension and/or lateral flexion and a positive Kemp's test.[14]

Piriformis Syndrome in Pregnancy

The piriformis muscle originates from the anterior sacrum and then crosses through the greater sciatic notch before attaching to the greater trochanter of the femur. The piriformis acts primarily as an external rotator when the hip is extended and adducts the hip when it is flexed. In pregnancy, the piriformis, along with other hip stabilizers, must compensate for the weakened gluteus medius and resultant lateral sway. Hypertrophy and hypertonicity result, and piriformis syndrome may develop. When the piriformis muscle is overused, irritated, or inflamed, it can lead to irritation of the adjacent sciatic nerve.[15] This is known clinically as piriformis syndrome.

Piriformis syndrome has been defined as a quartet of symptoms and signs: buttock pain aggravated on sitting, external tenderness near the greater sciatic notch, pain on any maneuver that increases piriformis muscle tension, and limitation of straight leg raising.[16] While piriformis syndrome is frequently underdiagnosed in the obstetric population, it should be suspected in any patient with symptoms of hip or sciatic pain.[17]

Category II Evaluation

Pelvic Sway:

One of the first components the practitioner will assess is pelvic sway.[18] Due to the pelvic instability commonly associated with the pregnant pelvis, a lateral or "side to side" sway is prevalent. This can manifest as restlessness while the patient is seated or shifting positions while standing. Historically the patient will report an inability to sit still or stand still.[18] In the acute phase of a Category II presentation, while standing, patients will present with a rapid or jerking correction to a proprioceptive center and in chronic cases, a presentation of muscle fatigue or discomfort will be notable. This presentation may present with gluteus medius weakness and/or a trendelenburg gait. Additionally, lateral pelvic sway can result in medial knee and lateral leg tenderness, also assessed as part of a complete SOT® clinical evaluation.

Rib head Tenderness:

With the patient seated or standing, the practitioner will palpate for quality and quantity of motion of the first rib/thoracic vertebra. Owing to pelvic destabilization and lateral shifting of forces, biomechanical compensation patterns are noted as cephalad as the cervicothoracic junction. In the acute phase of a Category II, the practitioner will note a hypermobility of the rib head articulation and in chronic cases, unilateral fixation is often present.[18] Palpatory tenderness is notable in all phases of dysfunction. It is important to note that in the spectrum of these cephalad dysfunctions, patients may present with scalene hypertonicity, thoracic outlet syndrome, glenohumeral dysfunction, or temporomandibular pain.

Arm Fossa Test:

This provocative test is neurologically reflexive in nature and is performed with the patient supine and with the practitioner visualizing the anterior pelvis in four functional quadrants. These quadrants are designated as upper and

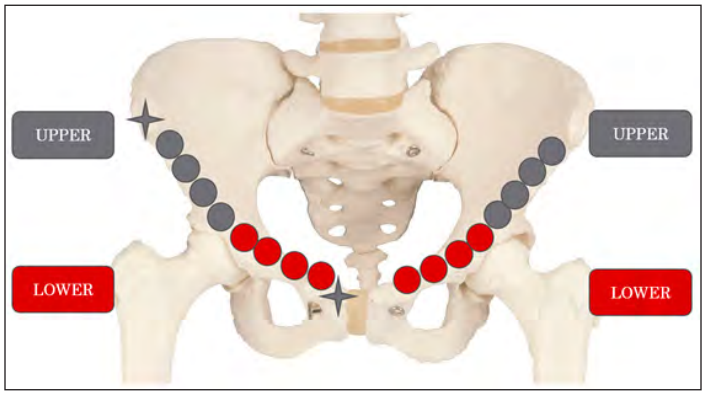

Figure 2: Four Functional Fossae.

lower fossae of the pelvis bordered superiorly by the anterior superior iliac crest and inferiorly by the pubic symphysis (Figure 2). While standing on the ipsilateral side and with the patient's arm raised ninety degrees to their trunk, the practitioner applies a quick and even four finger pressure to each fossa and thereby stimulating the inguinal ligament.[19] As this pressure is applied, the patient's raised arm is simultaneously challenged in the caudal direction with a synchronized burst of light pressure. Also synchronized with these actions, the practitioner delivers an auditory stimulus in the form of a command such as, "hold" (Figure 3). The patient is instructed to resist the light burst of caudal arm pressure and maintain their arm at ninety degrees. All four functional quadrants are assessed with this procedure. This test is noted as positive if a physical lag is noted in the patient's response to the reflexive challenge in any number of the functional fossae and suggests the presence of a Category II pelvic dysfunction. It is important to note that muscle strength is not being assessed, but rather, the speed of the patient's response.[19]

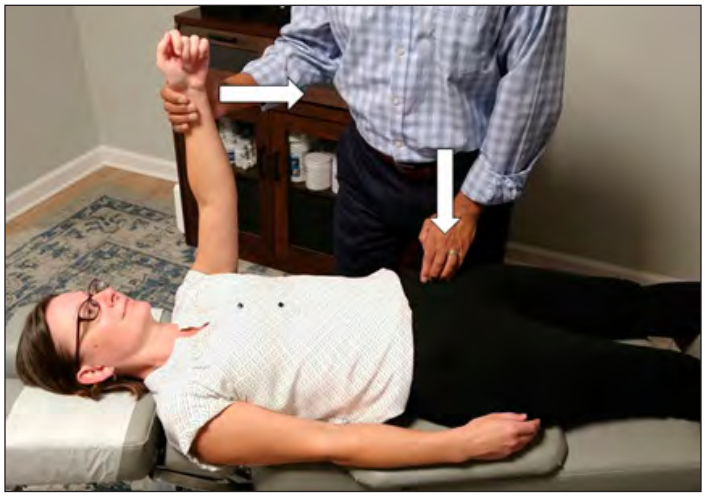

Figure 3: Arm Fossa Test.

Corrective Blocking:

Once the presence of a Category II dysfunction has been established, corrective pelvic blocking is indicated. If indicated, as per SOT® methodology, cervical subluxations are also to be corrected in conjunction with pelvic blocking. This allows cephalad dysfunctions to better respond to the pelvic correction.[19] The practitioner must then assess for the postural leg length deficiency in the supine position as this directs the placement of the corrective blocks. A table board should be placed under the patient's pelvis to provide an even foundation for the corrective blocks. On the side of leg length deficiency, a block is placed 90 degrees to the sagittal plane, 50% under the iliac crest and 50% under the lower lumbar musculature. On the contralateral side, a block is placed obliquely under the acetabulum, aimed at the medial edge of the opposing block (Figures 4a-c).[19] All four functional fossae are retested while the patient is actively being blocked and the procedure is terminated when the arm fossa test is negative bilaterally.

Figure 4a: Corrective blocking on the side of leg length deficiency. Block is placed 90 degrees to the sagittal plane.

Figure 4b: Corrective blocking on the contralateral side. Block is placed obliquely under the acetabulum, aimed at the medial edge of the opposing block.

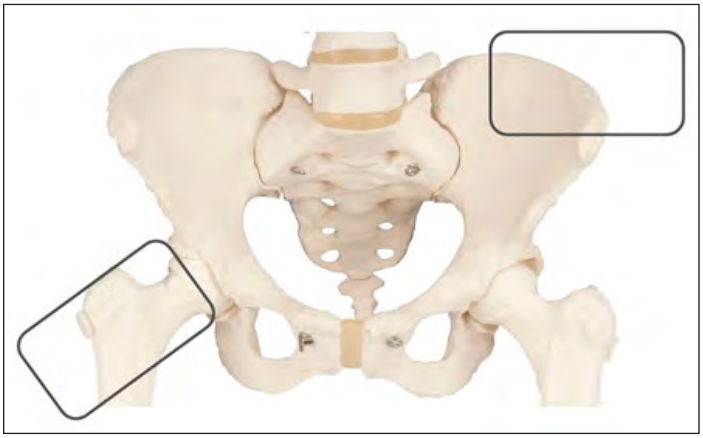

Figure 4c: Corrective Blocking Schematic.

Soft Tissue Corrections:
As an integral ancillary, facilitation of the tensor fascia latae and the iliopsoas musculature are to be addressed. A cross friction method referenced as the ilio-femoral roll procedure is utilized to ease tension in the hypertonic tensor fascia latae and sustained pressure upon the hypertonic iliacus/psoas musculature is employed as indicated.

Summary
As patients navigate the biomechanical changes associated with pregnancy, they may seek chiropractic care with the hope of being more comfortable and experiencing a healthy birth. It is important that practitioners are knowledgeable and proficient in technique approaches that cater to their patients' expectations while being gentle, safe, and effective. Understanding the biomechanical complexity of the lumbopelvic region and its stabilizers through pregnancy is imperative. With knowledge of SOT® and Category II presentations, practitioners are better positioned to improve structural and functional outcomes for their patients throughout pregnancy and birth.

Acknowledgements
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References:

1. Heese N. Major Bertrand DeJarnette: Six Decades of Sacro Occipital Research, 1924-1984. *Chiropractic History*. Jun 1991;11(1): 13-5.

2. Andrew CG. Considering non-optimal fetal positioning and pelvic girdle dysfunction in pregnancy: Increasing the available space. *J Clin Chiropr Pediatr*. 2010 Dec;11(2):783-788.

3. Berber MA, Satilmis G. Characteristics of Low Back Pain in Pregnancy, Risk Factors, and Its Effects on Quality of Life. *Pain Manag Nurs*. 2020;21(6):579-586.

4. Katonis P, Kampouroglou A, Aggelopoulos A, Kakavelakis K, Lykoudis S, Makrigiannakis A, Alpantaki K. Pregnancy-related low back pain. *Hippokratia*. 2011 Jul;15(3):205-10.

5. Sabino J, Grauer JN. Pregnancy and low back pain. *Curr Rev Musculoskelet Med*. 2008;1:137—141.

6. Bewyer KJ, Bewyer DC, Messenger D, Kennedy CM. Pilot data: association between gluteus medius weakness and low back pain during pregnancy. *Iowa Orthop J*. 2009;29:97—99.

7. Mueller MJ, et al. Tissue adaptation to stress: a proposed "physical stress theory" to guide physical therapy practice, education, and research. *Physical Therapy*. 2002;82(4):383—403.

8. Shah A, Bordoni B. Anatomy, bony pelvis and lower limb, gluteus medius muscle. StatPearls. Treasure Island (FL): StatPearls Publishing; 2022 Jan [Updated 2022 Jan 25] https://www.ncbi.nlm.nih.gov/books/NBK557509/. Accessed August 21, 2022.

9. Foti T, et al. A biomechanical analysis of gait during pregnancy. *The Journal of Bone and Joint Surgery*. 2000 May;82A(5):625—632.

10. Bordoni B, Varacallo M. Anatomy, bony pelvis and lower limb, iliopsoas muscle. StatPearls. Treasure Island (FL): StatPearls Publishing; 2022 Jan [Updated 2021 Jul 21] https://www.ncbi.nlm.nih.gov/books/NBK531508/. Accessed August 21, 2022.

11. Anderson CN. Iliopsoas: pathology, diagnosis, and treatment. *Clin Sports Med*. 2016 Jul;35(3):419-433.

12. Perolat R, Kastler A, Nicot B, Pellat JM, Tahon F, Attye A, Heck O, Boubagra K, Grand S, Krainik A. Facet joint syndrome: from diagnosis to interventional management. *Insights Imaging*. 2018 Oct;9(5):773-789.

13. Alexander CE, Sandean DP, Varacallo M. Lumbosacral facet syndrome. StatPearls. Treasure Island (FL): StatPearls Publishing; 2022 Jan [Updated 2022 May 1] https://www.ncbi.nlm.nih.gov/books/NBK441906/. Accessed August 21, 2022.

14. Hestbaek, L., Kongsted, A., Jensen, T.S. et al. The clinical aspects of the acute facet syndrome: results from a structured discussion among European chiropractors. *Chiropr Man Therap* 17, 2 (2009).

15. Hicks BL, Lam JC, Varacallo M. Piriformis syndrome. StatPearls. Treasure Island (FL): StatPearls Publishing; 2022 Jan [Updated 2022 Apr 21] https://www.ncbi.nlm.nih.gov/books/NBK448172/. Accessed August 21, 2022.

16. Hopayian, K., Danielyan, A. Four symptoms define the piriformis syndrome: an updated systematic review of its clinical features. *Eur J Orthop Surg Traumatol* 28, 155—164 (2018).

17. Sivrioglu AK, Ozyurek S, Mutlu H, Sonmez G. Piriformis syndrome occurring after pregnancy. *BMJ Case Rep*. 2013;2013:bcr2013008946. Published 2013 Mar 26. doi:10.1136/bcr-2013-008946.

18. Sacro Occipital Technique 1984. Major Bertrand DeJarnette, Nebraska City, NE 1984.

19. Monk R, 2006 SOT® Manual, SOT® O-USA: Winston-Salem, NC, 2006.

Review of:
Forces Involved with Labor and Delivery–A Biomechanical Perspective

Reviewed by:
Sue A. Weber DC, MSc Chiropractic Pediatrics

Original article by: MICHELE J. GRIMM
Annals of Biomedical Engineering, Vol. 49, No. 8, August 2021 (2021) pp. 1819—1835
https://pubmed.ncbi.nlm.nih.gov/33432512/

The purpose is to review the body of research focusing on the biomechanics of labor and delivery. It describes the forces generated during the different phases of parturition, specifically, how they affect the uterus and are transferred to the fetus. It highlights what can commonly happen to delay progression of labor and delivery.

This review encompasses studies as far back as the 1800's giving detail to the what we know about the process of contractions and the difficulty in measuring them for a biomechanical model. It describes the differences observed between forces generated during primiparous and multiparous deliveries. One specific maternal position is mentioned which is used to assist delivery by increasing forces. Two prevalent maternal systemic conditions which have a negative impact on labor and delivery are evaluated. Clinician applied forces including manual and instrumental traction are compared to the natural forces of labor. These forces are difficult to measure and are not consistent as each birth is unique. There is a discussion of how interns are trained manually in delivery and how gaining a feel for adequate force during traction is difficult.

As a review it articulates and discusses the material well and ends by giving some recommendations for studies which may provide valuable information for clinicians. It addresses where there are gaps in the literature and why there are difficulties gathering certain key bits of information. The review goes into detail about the factors which can complicate parturition. This could be better highlighted in the abstract as a major point. The conclusion points out that there is a significant complication rate during delivery, particularly during clinician assisted delivery. This could be mentioned in the abstract to make the purpose of the study clearer.

Review of:
Efficacy of pediatric integrative manual therapy in positional plagiocephaly: a randomized controlled trial

Reviewed by
Sue A. Weber DC, MSc Chiropractic Pediatrics

Original article by: Pastor-Pons I, Lucha-López MO, Barrau-Lalmolda M, Rodes-Pastor I, Rodríguez-Fernández ÁL, Hidalgo-García C, Tricás-Moreno JM.
Ital J Pediatr. **2021 Jun 5;47(1):132. doi: 10.1186/s13052-021-01079-4. PMID: 34090515; PMCID: PMC8180102.**
Text Link: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC8180102/ (accessed 29/6/2022)

The abstract summarizes the articles major points of the study.

The background gives a detailed description of risk factors for developing positional plagiocephaly (PP) as well as the goals of treatment.

This is an area of study in its infancy and well-designed studies are needed to assess the effects of manual therapy on PP.

In the measured parameters, there is no consideration of the inferior to superior length of the cranium. This is particularly affected in the breech baby where there is a relative flattening of the cranium.

It would be interesting to know what the physiotherapists who are conducting the study have for education within the field of cranial work for infants. There are no details regarding their competence other than their extended study. The authors articulate well the purpose of their study and maintain cohesion throughout. The title is concise and appropriate.

The authors make the statement that it is hypothesized that pediatric manual therapy is more effective in improving plagiocephaly than repositioning the infant complemented with sensory/motor training. This is not referenced as it would be interesting to know who is hypothesizing? What is the motivation for intervention? This is not mentioned and seems necessary to outline the goals of treatment by detailing the benefits of reducing the asymmetry caused by plagiocephaly. The author assumes here that that there is a benefit to treatment.

The design and methods the authors use work well to see the changes following a treatment period comparing the control and treatment group. The methods describe the basic concepts of the techniques used and refer to original texts for reference. These may not be available for everyone therefore it may make it difficult to duplicate the study.

The statistical methods seem appropriate, and there do not appear to be errors in calculations or repeated data. The literature cited supports the statements made by the authors.

As a randomized control study, the parameters are narrow to measure a specific change so not all the cranial and facial distortions of plagiocephaly are included. This is cited as a limitation. Perhaps more of these can be addressed in future studies.

Manual therapy is being compared to helmet therapy and what is not mentioned is that the manual therapy was one treatment session a week for a period of 10 weeks while the helmet therapy is usually one year for 22 hours a day. The helmet is not always tolerated due to skin reactions, it is quite warm during the summer and often develops an offensive odor. There are challenges to complying 100 percent with the helmet. An area of future study would be to instruct parents on one or two molding techniques to be performed at home for a 10-week period in combination with the weekly visits to see if this influenced outcomes.

This is an important study as it addresses a conservative aspect of care for a problem which has become more prevalent due to the back to sleep campaign.

JOURNAL ABSTRACTS

Click on the icon to read the full abstract or article.

New insights into the neural network of the nongravid uterus
Tanos V, Laidlaw J, Tanos P, Papadopoulou A.
Adv Clin Exp Med 2022 Jun 29.
doi: 10.17219/acem/149913. Epub ahead of print. PMID: 35766895.

Abstract
The human uterus is exposed to epigenetic factors during maturation, which might influence its neural network. The mesh muscle is formed from the circular muscle during development and maturation, and it coordinates the longitudinal and circular muscle function. The uterus has an autonomous neural network with contractility and propagation patterns that determine its reproductive potential and health during pregnancy and delivery. Emerging knowledge on the uterine neural network and mesh muscle ultrastructure contributes to new ideas and solutions on the role of intrauterine pressure and distending fluid intravasation during hysteroscopy, and even allows for improving the operative techniques of myomectomy, adenoma cytoreductive surgery and metroplasty. Good health and well-being start from the in utero stage of life. Prenatal and antenatal care are of paramount importance to minimize the risks of malnutrition and pollutants, and foster a healthy uterus. Research regarding the neural network, function and contractility of the nongravid uterus is a new chapter in gynecology that provides significant information for a better understanding and early diagnosis and treatment of uterine pathologies and early pregnancy support.
Keywords: estrogen; myometrium; neurotransmitter; progesterone; uterus.

Forces Involved with Labor and Delivery-A Biomechanical Perspective
Grimm MJ.
Ann Biomed 2021 Aug;49(8):1819-1835. doi: 10.1007/s10439-020-02718-3. Epub 2021 Jan 11. PMID: 33432512

Abstract
Childbirth is a primarily biomechanical process of physiology, and one that engineers have recently begun to address in a broader fashion. Computational models are being developed to address the biomechanical effects of parturition on both maternal and fetal tissues. Experimental research is being conducted to understand how maternal tissues adapt to intrauterine forces near the onset of labor. All of this research requires an understanding of the forces that are developed through maternal efforts-both uterine contractions and semi-voluntary pushing-and that can be applied by the clinician to assist with the delivery. This work reviews the current state of knowledge regarding forces of labor and delivery, with a focus on macro-level biomechanics.
Keywords: Biomechanics; Birth.

Efficacy of pediatric integrative manual therapy in positional plagiocephaly: a randomized controlled trial
Pastor-Pons I, Lucha-López MO, Barrau-Lalmolda M, Rodes-Pastor I, Rodríguez-Fernández ÁL, Hidalgo-García C, Tricás-Moreno JM.
Ital J Pediatr 2021 Jun 5;47(1):132. doi: 10.1186/s13052-021-01079-4. PMID: 34090515; PMCID: PMC8180102.

Abstract
Background: Positional plagiocephaly frequently affects healthy babies. It is hypothesized that manual therapy tailored to pediatrics is more effective in improving plagiocephalic cranial asymmetry than just repositioning and sensory and motor stimulation. **Methods:** Thirty-four neurologically healthy subjects aged less than 28 weeks old with a difference of at least 5 mm between cranial diagonal diameters were randomly distributed into 2 groups. For 10 weeks, the pediatric integrative manual therapy (PIMT) group received manual therapy plus a caregiver education program, while the controls received the same education program exclusively. Cranial shape was evaluated using anthropometry; cranial index (CI) and cranial vault asymmetry index (CVAI) were calculated. Parental perception of change was assessed using a visual analogue scale (-10 cm to +10 cm). **Results:** CVAI presented a greater decrease in PIMT group: 3.72±1.40% compared with 0.34±1.72% in the control group (p = 0.000). CI did not present significant differences between groups. Manual therapy led to a more positive parental perception of cranial changes (manual therapy: 6.66±2.07 cm; control: 4.25±2.31 cm; p = 0.004). **Conclusion:** Manual therapy plus a caregiver education program improved CVAI and led to parental satisfaction more effectively than solely a caregiver education program.

Trial registration number: NCT03659032; registration date: September 1, 2018. Retrospectively registered.

Keywords: Positional Plagiocephaly, Deformational Plagiocephaly, Manual therapy, Physical therapy
https://www.ncbi.nlm.nih.gov/pmc/articles/PMC8180102/

JOURNAL ABSTRACTS

Click on the icon to read the full abstract or article.

Effects of an Exercise Program on Brain Health Outcomes for Children With Overweight or Obesity: The ActiveBrains

Ortega FB, Mora-Gonzalez J, Cadenas-Sanchez C, et al.

Randomized Clinical Trial. *JAMA Netw Open.* 2022;5(8):e2227893. doi: 10.1001/jamanetworkopen.2022.27893.

Key Points

Question: Can an exercise intervention of aerobic plus resistance training improve cognitive and brain health outcomes for children with overweight or obesity? **Findings:** In this randomized clinical trial of 109 participants, exercise significantly improved intelligence and cognitive flexibility among preadolescent children with overweight or obesity. There was also a positive, smaller-magnitude significant effect of exercise on academic performance but no significant effect on inhibition and working memory or on structural and functional brain outcomes studied. **Meaning:** This study suggests that exercise can positively affect intelligence and cognitive flexibility during a sensitive period of brain development in childhood and, to a smaller extent, academic performance, indicating that an active lifestyle before puberty may lead to more successful life trajectories.

Abstract

Importance: Pediatric overweight and obesity are highly prevalent across the world, with implications for poorer cognitive and brain health. Exercise might potentially attenuate these adverse consequences. **Objectives:** To investigate the effects of an exercise program on brain health indicators, including intelligence, executive function, academic performance, and brain outcomes, among children with overweight or obesity and to explore potential mediators and moderators of the main effects of exercise. **Design, Setting, and Participants:** All preexercise and postexercise data for this 20-week randomized clinical trial of 109 children aged 8 to 11 years with overweight or obesity were collected from November 21, 2014, to June 30, 2016, with neuroimaging data processing and analyses conducted between June 1, 2017, and December 20, 2021. All 109 children were included in the intention-to-treat analyses; 90 children (82.6%) completed the postexercise evaluation and attended 70% or more of the recommended exercise sessions and were included in per-protocol analyses. **Interventions:** All participants received lifestyle recommendations. The control group continued their usual routines, whereas the exercise group attended a minimum of 3 supervised 90-minute sessions per week in an out-of-school setting. **Main Outcomes and Measures:** Intelligence, executive function (cognitive flexibility, inhibition, and working memory), and academic performance were assessed with standardized tests, and hippocampal volume was measured with magnetic resonance imaging. **Results:** The 109 participants included 45 girls (41.3%); participants had a mean (SD) body mass index of 26.8 (3.6) and a mean (SD) age of 10.0 (1.1) years at baseline. In per-protocol analyses, the exercise intervention improved crystallized intelligence, with the exercise group improving from before exercise to after exercise (mean z score, 0.62 [95% CI, 0.44-0.80]) compared with the control group (mean z score, —0.10 [95% CI, —0.28 to 0.09]; difference between groups, 0.72 SDs [95% CI, 0.46-0.97]; P < .001). Total intelligence also improved significantly more in the exercise group (mean z score, 0.69 [95% CI, 0.48-0.89]) than in the control group (mean z score, 0.07 [95% CI, —0.14 to 0.28]; difference between groups, 0.62 SDs [95% CI, 0.31-0.91]; P < .001). Exercise also positively affected a composite score of cognitive flexibility (mean z score: exercise group, 0.25 [95% CI, 0.05-0.44]; control group, —0.17 [95% CI, —0.39 to 0.04]; difference between groups, 0.42 SDs [95% CI, 0.13-0.71]; P = .005). These main effects were consistent in intention-to-treat analyses and after multiple-testing correction. There was a positive, small-magnitude effect of exercise on total academic performance (mean z score: exercise group, 0.31 [95% CI, 0.18-0.44]; control group, 0.10 [95% CI, —0.04 to 0.24]; difference between groups, 0.21 SDs [95% CI, 0.01-0.40]; P = .03), which was partially mediated by cognitive flexibility. Inhibition, working memory, hippocampal volume, and other brain magnetic resonance imaging outcomes studied were not affected by the exercise program. The intervention increased cardiorespiratory fitness performance as indicated by longer treadmill time to exhaustion (mean z score: exercise group, 0.54 [95% CI, 0.27-0.82]; control group, 0.13 [95% CI, —0.16 to 0.41]; difference between groups, 0.42 SDs [95% CI, 0.01-0.82]; P = .04), and these changes in fitness mediated some of the effects (small percentage of mediation [approximately 10%-20%]). The effects of exercise were overall consistent across the moderators tested, except for larger improvements in intelligence among boys compared with girls. **Conclusions and Relevance:** In this randomized clinical trial, exercise positively affected intelligence and cognitive flexibility during development among children with overweight or obesity. However, the structural and functional brain changes responsible for these improvements were not identified.

Trial Registration ClinicalTrials.gov Identifier: NCT02295072

Global Changes in Child and Adolescent Physical Activity During the COVID-19 Pandemic: A Systematic Review and Meta-analysis

Neville RD, Lakes KD, Hopkins WG, et al.

JAMA Pediatr 2022;176(9):886—894. doi:10.1001/jamapediatrics.2022.2313

Key Points

Question: To what extent has the COVID-19 pandemic affected the global physical activity levels of children and adolescents? **Findings:** In this systematic review and meta-analysis of 22 international longitudinal studies that included 14,216 children 18 years and younger, pooled estimates revealed a decrease of 17 minutes per day in children's moderate-to-vigorous physical activity from prepandemic to during the COVID-19 pandemic. **Meaning:** Restrictions implemented during the COVID-19 pandemic have affected children's levels of physical activity, particularly moderate-to-vigorous physical activity; children's movement behaviors should be at the forefront of pandemic recovery efforts.

Abstract

Importance: Numerous physical distancing measures were implemented to mitigate the spread of the COVID-19 virus, which could have negatively affected child and adolescent physical activity levels. **Objectives:** To conduct a systematic review and meta-analysis of the literature that used validated measures to document changes in child and adolescent physical activity during the COVID-19 pandemic and to estimate whether changes in physical activity differed between participant-level, contextual, and methodological moderators. **Data Sources:** PubMed, PsycInfo, SPORTDiscus, Web of Science, Scopus, CINAHL, and MEDLINE were searched (from January 1, 2020, to January 1, 2022). A total of 1085 nonduplicate records were retrieved. Study Selection Studies were included if they reported (1) changes in the duration of physical activity at any intensity for children or adolescents (age ≤18 years) comparing before and during the COVID-19 pandemic using validated physical activity measurement tools and were (2) from general population samples, (3) peer-reviewed, and (4) published in English. **Data Extraction and Synthesis:** A total of 126 articles underwent full-text review. Data were analyzed using a random-effects meta-analysis, which was conducted in January 2022. **Main Outcomes and Measures:** Change in the duration of engagement in physical activity at any intensity comparing before and during COVID-19. **Results:** Twenty-two studies including 46 independent samples and 79 effect sizes from 14,216 participants (median age, 10.5 years; range, 3-18 years) were included. The percentage change in the duration of engagement in total daily physical activity from before to during COVID-19 was -20% (90% CI, -34% to -4%). Moderation analyses revealed that changes were larger for higher-intensity activities (-32%; 90% CI, -44% to -16%), corresponding to a 17-minute reduction in children's daily moderate-to-vigorous physical activity levels. The reduction in physical activity was also larger for samples located at higher latitudes (37%; 90% CI, -1% to 89%) and for studies with a longer duration between physical activity assessments (25%; 90% CI, -0.5% to 58%). **Conclusions and Relevance:** Children and adolescents have experienced measurable reductions in physical activity during the COVID-19 pandemic. Findings underscore the need to provide bolstered access to support and resources related to physical activity to ensure good health and social functioning among children and adolescents during pandemic recovery efforts.

Using Time-out for Child Conduct Problems in the Context of Trauma and Adversity: A Nonrandomized Controlled Trial

Roach AC, Lechowicz M, Yiu Y, Mendoza Diaz A, Hawes D, Dadds MR.

JAMA Netw Open, 2022;5(9):e2229726. doi:10.1001/jamanetworkopen.2022.29726.

Key Points

Question: Are parenting programs that include time-out less effective or even harmful for children exposed to adverse childhood experiences? **Findings:** In this nonrandomized clinical trial of 205 families, children with conduct problems and high adversity exposure experienced equivalent, if not greater, outcomes, measured by the parent-reported Strengths and Difficulties Questionnaire, after a parenting program that included time-out, compared with children with low adversity exposure. **Meaning:** This study suggests that, despite concerns that time-out is contraindicated for children who have experienced adversity, parenting programs that include time-out appear to be beneficial for children with or without adversity exposure for management of emotional and behavioral difficulties.

Abstract

Importance: Exposure to adverse childhood experiences substantially increases the risk of chronic health problems. Originally designed to treat child conduct problems, parent management training programs have been shown to be effective in preventing children from being exposed to further adversity and supporting children's recovery from adversity; however, there are increasing concerns that a core component of these programs, the discipline strategy time-out, may be harmful for children with a history of exposure to adversity. **Objective:** To investigate the comparative benefits and potential harms to children exposed to adversity that are associated with parenting programs that include time-out. **Design, Setting, and Participants:** This nonrandomized waiting list—controlled

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clinical study was conducted at a specialist clinic for the treatment of conduct problems in Sydney, Australia. The self-referred sample included children with conduct problems and their caregivers. Eligibility was confirmed through clinician-administered interviews. Data were collected between February 14, 2018, and February 1, 2021. **Interventions:** Caregivers participated in a 10-session, social learning—based parent management training program. Caregivers were provided with parenting strategies aimed at encouraging desired behaviors through effective reinforcement and managing misbehavior through consistent limit setting, including the use of time-outs. **Main Outcomes and Measures:** The primary outcome was the parent-reported Strengths and Difficulties Questionnaire score, and secondary outcomes included subscale scores from the clinician-administered Diagnostic Interview Schedule for Children, Adolescents, and Parents. Multi-informant measures of child adversity were collected using the parent-reported Adverse Life Experiences Scale and the clinician-rated Maltreatment Index. **Results:** A total of 205 children were included in analysis (156 in the full intervention and 49 in the control condition; 158 boys [77.1%]; mean [SD] age, 5.6 [1.8] years [range, 2-9 years]). Compared with children with low adversity exposure, children with high adversity exposure showed greater reductions in the Strengths and Difficulties Questionnaire score from baseline (mean difference, 3.46 [95% CI, 1.51-5.41]; P < .001) to after treatment (mean difference, 1.49 [95% CI, -0.46 to 3.44]; P = .13) and in the internalizing symptom subscale score (baseline mean difference, 1.00 [95% CI, -2,00 to 0.00]; P = .50; posttreatment mean difference, 0.06 [95% CI, -0.82 to 0.94]; P = .90). No significant differences in the externalizing symptom subscale score were found. **Conclusions and Relevance:** In this nonrandomized clinical study, children with high exposure to adversity experienced equivalent, if not greater, benefits associated with parenting programs that include time-out compared with children with low exposure to adversity. Results suggest that time-out was an effective component of parenting programs for children exposed to adversity.

Trial Registration: anzctr.org.au Identifier: ACTRN12617001472369

Developmental Variability in Autism Across 17 000 Autistic Individuals and 4000 Siblings Without an Autism Diagnosis Comparisons by Cohort, Intellectual Disability, Genetic Etiology, and Age at Diagnosis

Susan S. Kuo, PhD; Celia van der Merwe, PhD; Jack M. Fu, PhD; et alCaitlin E. Carey, PhD; Michael E. Talkowski, PhD; Somer L. Bishop, PhD; Elise B. Robinson, DSc.

JAMA Pediatr Published online July 18, 2022. doi:10.1001/jamapediatrics.2022.2423.

Abstract

Importance: Presence of developmental delays in autism is well established, yet few studies have characterized variability in developmental milestone attainment in this population. **Objective:** To characterize variability in the age at which autistic individuals attain key developmental milestones based on co-occurring intellectual disability (ID), presence of a rare disruptive genetic variant associated with neurodevelopmental disorders (NDD), age at autism diagnosis, and research cohort membership. **Design:** The study team harmonized data from 4 cross-sectional autism cohorts: the Autism Genetics Research Exchange (n = 3284; 1997-2015), The Autism Simplex Collection (n = 694; 2008-2011), the Simons Simplex Collection (n = 2753; 2008-2011), and the Simons Foundation Powering Autism Research for Knowledge (n = 10 367; 2016-present). The last sample further included 4,145 siblings without an autism diagnosis or ID. **Participants:** Convenience sample of 21,243 autistic individuals or their siblings without an autism diagnosis aged 4 to 17 years. **Main Outcomes and Measures:** Parents reported ages at which participants attained key milestones including smiling, sitting upright, crawling, walking, spoon-feeding self, speaking words, speaking phrases, and acquiring bladder and bowel control. A total of 5,295 autistic individuals, and their biological parents, were genetically characterized to identify de novo variants in NDD-associated genes. The study team conducted time-to-event analyses to estimate and compare percentiles in time with milestone attainment across autistic individuals, subgroups of autistic individuals, and the sibling sample. **Results:** Seventeen thousand ninety-eight autistic individuals (mean age, 9.15 years; 80.8% male) compared with 4,145 siblings without autism or ID (mean age, 10.2 years; 50.2% female) showed delays in milestone attainment, with median (IQR) delays ranging from 0.7 (0.3-1.6) to 19.7 (11.4-32.2) months. More severe and more variable delays in autism were associated with the presence of co-occurring ID, carrying an NDD-associated rare genetic variant, and being diagnosed with autism by age 5 years. More severe and more variable delays were also associated with membership in earlier study cohorts, consistent with autism's diagnostic and ascertainment expansion over the last 30 years. **Conclusions and Relevance:** As the largest summary to date of developmental milestone attainment in autism, to our knowledge, this study demonstrates substantial developmental variability across different conditions and provides important context for understanding the phenotypic and etiological heterogeneity of autism.

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At a Crossroads–Reconsidering the Goals of Autism Early Behavioral Intervention From a Neurodiversity Perspective

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The neurodiversity perspective posits that each person has a unique brain and a unique combination of traits and abilities and asserts that many challenges faced by autistic individuals stem from a lack of fit between the characteristics of autistic people and society's expectations and biases. The neurodiversity movement is akin to a civil rights movement. Among its goals are reducing stigma, increasing accessibility, and ensuring that autistic individuals' voices are represented in decisions about autism research, policy, and clinical practice. The neurodiversity movement is having a growing influence on the scientific community, as evidenced in the recent pause in a large autism genetic study based on concerns raised by the autism community.[1] It is also affecting autism practitioners as, increasingly, parents are expressing reservations about enrolling their child in early intervention programs, citing concerns that such programs do not value neurodiversity and, instead, prioritize changing their child's behavior to fit neurotypical norms.

The diagnostic odyssey of autism: a cross-sectional study of 3 age cohorts of children from the 2016—2018 National Survey of Children's Health

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PMID: 34629109 PMCID: PMC8504038 doi: 10.1186/s13034-021-00409-y.

Abstract

Background: Autism prevalence has increased rapidly in recent years, however, nationally representative estimates on the ages of first identification and intervention are out of date. Objectives: (1) To estimate the ages at which children with autism receive their first diagnosis, intervention plan, and developmental services; and (2) To evaluate differences in ages at events by birth cohort and sociodemographic characteristics. **Methods:** Using cross-sectional data from the 2016—2018 National Survey of Children's Health (NSCH), we examined associations via linear regression among a sample of 2303 children aged 2—17 years old, who had ever been diagnosed with autism and either (1) ever had a plan for special education or early intervention, or (2) ever received special services to meet developmental needs. Exposures included age cohort, child, household and healthcare provider characteristics. **Results:** Most children in the study sample (n = 2303) were over age 6 years, male, of non-Hispanic white race ethnicity and had mild/moderate autism. Mean ages (years) at first diagnosis was 4.56 (SE = 0.13); first plan was 4.43 (SE = 0.11); and first services was 4.10 (SE = 0.11). After adjustment for exposures and survey year, the middle childhood cohort was 18 months older at first intervention (ß = 1.49, 95% CI, 1.18—1.81), and adolescents were 38 months older at first diagnosis (ß = 3.16, 95% CI, 2.72—3.60) compared to those in early childhood. Younger ages at events were observed among: Hispanic/Latinx as compared to white children, those with moderate or severe symptoms as compared to mild symptoms, and children who received their diagnosis from a specialist as compared to psychologists or psychiatrists. **Conclusions:** Children with autism receive their first diagnosis, intervention plans and developmental services at younger ages than they had in the past. Future research is needed to identify the mechanisms for these improvements in early identification and intervention to accelerate additional progress.

JOURNAL ABSTRACTS

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Defining the anatomy of the neonatal lingual frenulum
Mills N, Keough N, Geddes DT, Pransky SM, Mirjalili SA.
Clin Anat. 2019 Sep;32(6):824-835. doi: 10.1002/ca.23410. Epub 2019 Jun 3. PMID: 31116462.

Abstract
The lingual frenulum is recognized as having the potential to limit tongue mobility, which may lead to difficulties with breastfeeding in some infants. There is extensive variation between individuals in the appearance of the lingual frenulum but an ambiguous relationship between frenulum appearance and functional limitation. An increasing number of infants are being diagnosed with ankyloglossia, with growing uncertainty regarding what can be considered "normal" lingual frenulum anatomy. In this study, microdissection of four fresh tissue premature infant cadavers shows that the lingual frenulum is a dynamic, layered structure formed by oral mucosa and the underlying floor of mouth fascia, which is mobilized into a midline fold with tongue elevation and/or retraction. Genioglossus is suspended from the floor of mouth fascia, and in some individuals can be drawn up into the fold of the frenulum. Branches of the lingual nerve are located superficially on the ventral surface of the tongue, immediately beneath the fascia, making them vulnerable to injury during frenotomy procedures. This research challenges the longstanding belief that the lingual frenulum is a midline structure formed by a submucosal "band" or "string" and confirms that the neonatal lingual frenulum structure replicates that recently described in the adult. This article provides an anatomical construct for understanding and describing variability in lingual frenulum morphology and lays the foundation for future research to assess the impact of specific anatomic variants of lingual frenulum morphology on tongue mobility.

Clin. Anat. 32:824-835, 2019.

Keywords: ankyloglossia; fascia; floor of mouth; frenotomy; lingual frenulum; neonatal; neonate; surgery; tongue tie.

Osteopathic Manipulative Treatment Decreases Hospital Stay and Healthcare Cost in the Neonatal Intensive Care Unit
Roland H, Brown A, Rousselot A, Freeman N, Wieting JM, Bergman S, Mondal D.
Medicines. 2022; 9(10):49. https://doi.org/10.3390/medicines9100049.

Abstract
Osteopathic manipulative treatment (OMT) is used in both inpatient and outpatient settings. Evidence suggests that OMT can reduce both patients' recovery time and the financial cost of their acute medical treatment and rehabilitation. Multiple studies from neonatal intensive care units (NICUs) are presented in this article that demonstrate infants treated with OMT recover faster, are discharged earlier, and have lower healthcare costs than their non-OMT-treated counterparts. Data clearly show that adjunctive OMT facilitates feeding coordination in newborns, such as latching, suckling, swallowing, and breathing, and increases long-term weight gain and maintenance, which reduces hospital length of stay (LOS). Osteopathic techniques, such as soft tissue manipulation, balanced ligamentous tension, myofascial release, and osteopathic cranial manipulation (OCM), can reduce regurgitation, vomiting, milky bilious, or bloody discharge and decrease the need for constipation treatment. OMT can also be effective in reducing the complications of pneumonia in premature babies. Studies show the use of OCM and lymphatic pump technique (LPT) reduces the occurrence of both aspiration and environmentally acquired pneumonia, resulting in significantly lower morbidity and mortality in infants. Based on published findings, it is determined that OMT is clinically effective, cost efficient, a less invasive alternative to surgery, and a less toxic choice to pharmacologic drugs. Therefore, routine incorporation of OMT in the NICU can be of great benefit in infants with multiple disorders. Future OMT research should aim to initiate clinical trial designs that include randomized controlled trials with larger cohorts of infants admitted to the NICU. Furthermore, a streamlined and concerted effort to elucidate the underlying molecular mechanisms associated with the beneficial effects of OMT will aid in understanding the significant value of incorporating OMT into optimal patient care.

Keywords: neonatal intensive care unit; NICU; osteopathic medicine; osteopathic manipulative treatment; hospital stay; healthcare cost.