JCCP

JOURNAL OF CLINICAL CHIROPRACTIC PEDIATRICS

VOLUME 22 • NO. 1 • JUNE 2023
PUBLICATION OF THE COUNCIL ON CHIROPRACTIC PEDIATRICS
INTERNATIONAL CHIROPRACTORS ASSOCIATION

JCCP Journal of Clinical Chiropractic Pediatrics

The Journal of Clinical Chiropractic Pediatrics (JCCP) is the official peer-reviewed journal of the Council on Chiropractic Pediatrics, 6400 Arlington Boulevard, Suite 800, Falls Church, Virginia 22042, USA.

Editorial Correspondence: Correspondence should be sent to:
Editor, JCCP
ICA Council on Chiropractic Pediatrics
6400 Arlington Blvd., Suite 800, Falls Church, Virginia 22042, USA
Email: pediatricscouncil@chiropractic.org or svallonedc@aol.com

TABLE OF CONTENTS

JCCP JOURNAL OF CLINICAL CHIROPRACTIC PEDIATRICS

VOLUME 22, NUMBER 1 JUNE 2023

Publishing Offices:
ICA Council on Chiropractic Pediatrics
6400 Arlington Boulevard, Suite 800, Falls Church, Virginia 22042 U.S.A.

GUIDELINES FOR AUTHORS

JCCP JOURNAL OF CLINICAL CHIROPRACTIC PEDIATRICS

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

The following will be considered:

Case Reports and Case Series – presentations of individual or groups of cases deemed to be of interest to the professional and scholarly community.

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

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

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

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

Guidelines for submission

All manuscripts are accepted purely for consideration. They must be original works and should not be under consideration by any other journal or publisher at the time of submission. They must be accompanied by a TRANSFER OF COPYRIGHT form, signed by all authors and by the employer if the paper is the result of a "work for hire." It is understood that while the manuscript is under consideration it will not be sent to any other publication. In the case of multiple authors, a transmittal letter should designate one author as correspondent.

Manuscripts may be sent to editor at svallonedc@aol.com. Manuscript should be in document style MS Word (or compatible) and unformatted. PDFs will not be accepted.

The paper must include an abstract or summary. This abstract/summary should state the purpose of the paper (objective), procedures, methods, main findings (results) and principal conclusions. Also, any key words or phrases that will assist indexers should be provided.

References must be cited for all materials derived from the works of other people and previously published works. Reference numbers in superscript must be assigned in the order of citation in the paper.

Tables – Each table or figure should be on a separate page and not imbedded in the manuscript. If the table is from another publication, permission to publish must be granted and the publication acknowledged.

Photographs – Photographs may be in color or in grayscale and scanned at 300 dpi with sharp contrast. Patient photographs must have consent form signed by the individual or parent or guardian in the case of a minor.

Informed Consent – If the research/study involves experimental investigations performed on humans the manuscript must include a statement that informed consent was obtained from the individuals involved in the investigation.

Patient Anonymity – Patient names or any information that could identify a specific patient should be avoided. All case reports, with or without identifying photographs accompanying a manuscript must have a consent form signed by the individual or parent or guardian in the case of a minor. These are to include any requests for blocking faces, etc.

Acknowledgements – Any illustrations from other publications must be acknowledged. It is the author's responsibility to obtain written permission from the publisher and/or author for their use.

All manuscripts deemed appropriate for publication by the editor will be sent blind to at least two reviewers. If the manuscript is accepted, the author will be notified. If substantive changes are required, the paper will be returned to the author and the author must re-submit a clean copy of the revised manuscript. Author will be given a tentative date for publication if accepted. Manuscripts not accepted for publication will be returned to the author without comment.

Instructions to Authors – Summary

See *Uniform Requirements for Manuscripts Submitted to Biomedical Journals* for detailed information http://www.icmje.org/.

General formatting guidelines

- All submission components must be submitted electronically.
- Only manuscripts in English are accepted.
- Submit manuscripts as **Microsoft Word** documents.
- Use 1" margins on all sides
- Use Arial 12 point black font
- Capitalize only the first letter in the title, and any proper nouns.
- Do not justify text.
- Do not use column function
- Number all pages at bottom right.
- Double-space manuscript. Single-space references, tables or figure legends.
- Do not abbreviate words or terms the first time they are introduced; at that time, provide the abbreviation in parentheses and use it from that point forward.
- Number citations consecutively using superscripted Arabic numerals and place all references in a **Reference** section immediately at the end of your section.
- Run spell check and grammar check after completing the manuscript. Use American English spelling and units of measurement.

Submission Components

- ***JCCP* authorship form**—submit separately from manuscript. All authorship forms may be combined in a single PDF. Each author must complete this form, scan and return it electronically to the editor before the manuscript can be processed.
- ***JCCP* Patient (or Parent/Guardian) Permission to Publish Form**—one form for each case (1 for case report; multiple individual forms for case series) — all forms may be combined as a single PDF.
- **Permission to acknowledge forms:** All individuals named in the Acknowledgements section of the manuscript must sign a permission form. The corresponding author may use his or her own form, or use the one *JCCP* provides–submit separately from manuscript. All permission forms may be combined as a single PDF.
- **Cover letter**—submit as separate document, either Word or PDF.

The following items MUST be submitted as a Word document.

Cover letter—Explain why your manuscript is appropriate for *JCCP*.

Document— Each of the following should be on a separate page. Use page break function to separate page, not repeated line breaks to get to a new page.

- Title page
- Abstract
- Manuscript
- Acknowledgements
- References
- Tables
- Figures

Title page

- Title of article—ONLY CAPITALIZE FIRST LETTER OF FIRST WORD
- Running head (limited to 40 characters)
- Word count (excluding references, tables and figures)
- Number of tables
- Number of figures
- Authors
  - Name, with all degrees (do not include Bachelor's level degrees)
  - Current title/position and affiliation, including city, state and country
- Corresponding author
  - Name
  - Mailing address, phone, fax
  - E-mail address; provide alternative e-mail address if possible

Abstract—not to exceed 250 words. It may be structured or unstructured. Structured abstracts usually include the following sections: Purpose, Methods (include study design in this section), Results, Conclusion. For case reports and case series, see document, "Instructions for Case Reports and Case Series."

Manuscript Components

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.
- Methods: describe the methods used to accomplish the study, in detail sufficient to allow the informed reader to evaluate their appropriateness.
- Results: present the results of the study, without interpretation.
- Discussion: describe limitations of the study; interpret results; compare results to those of other relevant studies; discuss value and implications of the study.
- Inclusion of appendices is discouraged.

Instructions to Authors – Summary

Tables

- Number tables consecutively in text, using Arabic numerals (1, 2, 3 etc.)
- Place each table on a separate page at the end of the section, immediately following the References section.
- Use "table" function in Word to construct tables; do NOT use tab or space keys to form columns and rows. Use table "normal" style to construct table. Do not insert vertical lines between columns; do not use grids. Place horizontal line under table title and at end of table, separating the table from any footnotes. You may place horizontal lines under headings in the table for clarity.
- 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.
- Use left-justification to align numbers in columns.

Figures

- Place figure title and legend on page with the figure.
- Figures must be submitted electronically. Acceptable file formats: DOC, JPG, PDF. Figures may be embedded at the end of the manuscript text file or loaded as separate files for submission purposes. Should not be imbedded within the manuscript text
- Hand-drawn illustrations are not acceptable.
- Provide documentation of permission for any figures that are not original.

Acknowledgements

Include a statement disclosing any funding support for the project or project personnel, or any other potential conflicts of interest. Acknowledge only individuals or organizations who provided input or resources to the project that were above and beyond their usual responsibilities. All individuals acknowledged must provide written permission to use their name; these permissions must accompany the manuscript at the time of submission (scan documents and submit electronically).

Reference format—examples

- *Journal article*: Jefferies LJ, Milanese SF, Grimmer-Somers KA. Epidemiology of adolescent spinal pain: A systematic overview. *Spine* 2007;32:2630-2637.
- Book: Task Force on Community Preventive Services. Guide to Community Preventive Services. New York: Oxford University Press; 2005.
- Website/webpages: Author. Title. Name of website. URL. Date of publication. Updated date (if applicable). Date accessed. Example: Fox F. Promoting and sustaining collaborative networks in pediatrics. Pew Research Center. http://www.pewinternet.org/2013/06/14/promoting-and-sustaining-collaborative-networks-in-pediatrics/. Published June 14, 2013. Accessed September 3, 2017.

Permission to acknowledge forms

All individuals named in the Acknowledgements section of the manuscript must sign a permission form. The corresponding author may use his or her own form, or use the one *JCCP* provides.

Title Page Format

Running Head:
Word count (excluding references, tables and figures):
Number of tables:
Number of figures:

Authors (in correct order)
Name, degrees
Current title/position and institution (if applicable)
City, State, Country

Corresponding Author
Name
Address
Phone Number:
Fax:
Email:

Journal of Clinical Chiropractic Pediatrics Authorship Form

Each author must read and sign the statements on 1) authorship responsibility and contribution, 2) financial disclosure and conflict of interest, 3) copyright transfer. **The corresponding author must sign the Acknowledgement Statement** and email the completed form to Svallonedc@aol.com to initiate manuscript processing.

Manuscript title: _____

1. Authorship Responsibility and Contribution

• I certify that this submission represents original work, and that neither this submission nor a substantially similar one has been published or is under consideration for publication elsewhere in any medium (paper or electronic). I also affirm that this submission is not subject to copyright or any other rights except those of the current authors.

• I certify that if so requested by the editor, I will provide the data or cooperate in obtaining the data on which this submission is based, for review by the journal's designated representative(s).

• I agree that the corresponding author may represent me to review proofs and make other decisions regarding the submission. I have approved the submission.

• I certify that I meet the criteria for authorship, having made substantive contribution to the manuscript as indicated below (check all that apply).

___ Development of project concept or hypothesis
___ Study design and development of methodology
___ Project implementation
___ Data collection and management
___ Data analysis and interpretation of results
___ Literature search and review
___ Manuscript writing
___ Other (specify contribution)_____

2. Financial Disclosure and Conflict of Interest

I certify that all sources of extramural support of this submission, and the role of any funding agencies in the conduct of the study have been clearly described in the Acknowledgements section of the submission.

Check one of the following two statements:

❑ I certify that I have no financial interests, relationships or affiliations related to the project or materials addressed in the submission.

OR

❑ I certify that any potential conflicts of interest, including financial interests, relationships or affiliations related to this submission are disclosed in the Acknowledgements section of the manuscript.

3. Copyright Transfer

In consideration of the action of the *Journal of Clinical Chiropractic Pediatrics* in reviewing and editing this submission (including manuscripts, tables, figures and any supplemental documents), I hereby transfer, assign, or otherwise convey all copyright ownership including all rights and incidental thereto, exclusively to the *Journal of Clinical Chiropractic Pediatrics*.

I also understand that if the manuscript is not accepted for publication by the *Journal of Clinical Chiropractic Pediatrics* I will be notified and the transfer of copyright will be null and void.

_____	_____	_____
Signature	e-mail address	date signed

Acknowledgement statement to be signed by corresponding author

All individuals named in Acknowledgements section should provide written permission. I certify that:

• All individuals who have made substantive contributions to the submission but who do not qualify as authors have been named, along with their specific contribution in the Acknowledgements.

• All individuals so named have provided me with their written permission to be named.

• If no Acknowledgement section is included in the submission, there are no other contributors to the manuscript.

_____	_____	_____
Corresponding Author Signature	e-mail address	date signed

Journal of Clinical Chiropractic Pediatrics

Patient Consent Form for Case Report

Print name:_____

If patient is a minor, print parent/guardian name: _____

I have read the information about me/minor and/or seen the photograph to be published.
I give my consent for this material to appear in a scientific journal.

I understand the following:
(1) My name/minor's name will not be attached to the material. The authors of the article will make every attempt to keep my identity/minor's identity anonymous. I understand, however, that they cannot guarantee complete anonymity. It is possible that someone, such as someone who works in this clinic or one of my relatives, might be able to identify me/minor.

(2) The material will only be published in a scientific journal.

(3) The material will not be used for advertising.

Signed:_____ Today's date: _____
(if patient is a minor, parent or guardian signs.)

Journal of Clinical Chiropractic Pediatrics

Permission to Acknowledge

I give my permission to be acknowledged in the manuscript,

_____

which is to be submitted to the *Journal of Clinical Chiropractic Pediatrics*.

_____ _____
Signature Date Signed

_____
Print Name

Instructions for Case Reports and Case Series

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)

Additional interesting articles to read about EBM and writing and EBCR:

Review an example of an EBCR at:
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

Editorial

Everybody paddle the canoes together!

Twice a year, it's a frantic rush to reel in the fishing lines cast out across various continents so another issue of the *JCCP* can be published with contributions from individuals who step out of their comfort zone, their daily workload, to author a manuscript that will hopefully offer some clinical insight, some cumulative data, some educational nugget to help each of us in our daily chiropractic practice, as an author, teacher, mentor or perhaps another healthcare provider or parent who is searching for the missing piece for their patient or client or their own family.

Chiropractic has been in the eye of the storm since its inception. Recently, I was honored to participate in an international forum to attempt to codify current practice guidelines for pediatric chiropractors to once more, attempt to represent what we do with accountability, clinical insight and experience combined with what research is available. It was a positive experience with contributions that were constructive and respectful until the final product was sculpted and set out to dry (and we still write to each other as we see things that could be tweaked, added, perhaps said more concisely… as practice, and life, are meant to be fluid!).

In comparison, though, part of what we've all realized is that more time and effort, let alone all the research money, is being spent producing literature criticizing our "lack of evidence" rather than our "wealth of positive clinical outcomes". And limiting all the meagre research funding to only one possible positive outcome of chiropractic (low back pain) leaves us without the opportunity to explore the "unproven" results obtained when applied appropriately in a myriad of different clinical scenarios.

Challenges to the profession and its scope of practice, spurred the formation of an international group spearheaded by two chiropractors in Australia whose colleagues have been under political scrutiny. In an ongoing email discussion between participants, my co-editor Dr. Cheryl Hawk (Texas Chiropractic College) and one of the founding members of the group, Dr. Lyndon Amorin-Woods (Murdoch University Chiropractic Clinic), shared these thoughts:

Dr. Hawk
"I keep thinking of a comment a colleague (psychologist, Bob Jensen) made years ago, that the purpose of research is to IMPROVE practice, not PROVE it. Chiropractic has had to spend so much time and money trying to PROVE that its practice is legitimate (which we've only done thoroughly for LBP, because the medical profession is able to understand "work on back = might help back pain") that we have very little left over to do research to IMPROVE practice. And the political opponents are twisting the research (that is, basing it on the "lack of research" equals "lack of effectiveness" fallacy)[1-3] to *disprove* it."

As Marson Smith stated: "Systematic reviews of healthcare interventions need to be as clear as the evidence will support. Many people will read only the abstracts of systematic reviews. Leaving readers with the impression that there is no difference between alternative treatments may result in dangerously misinformed clinical decisions and failure to address important uncertainties in additional research."[2]

Dr. Amorin Woods
The point is this. *It is meaningless to consider just the evidence of the 'intervention' without that for the options or alternatives.* 95% of healthcare is lacking evidence, spinal manipulative therapy (SMT) is no worse (or better) than most other medical or other interventions for most childhood conditions.

That is why, recognizing that the clinician is (virtually) always working within a context of clinical uncertainty, that the principle is to be 'defendable' rather than 'right' or definitive.

Of course, one must always strive to be 'correct' but must also recognize the limitations inherent in 'diagnosis' or 'clinical labelling'."

Always ahead of her time, Dr. Hawk wrote a paper in 1998 while at the Palmer Center for Chiropractic research clearly stating that we are a profession not a procedure.

"…its application (the chiropractic adjustment) must be informed by a unique approach to healing and health that is distinct from, although not necessarily at odds with, the medical model. For it is the philosophy, the intuitive knowledge - the belief system - that differentiates a complete system from a procedure."

So, here we are. Some of us, clinicians in the field, some researchers in the "lab" and some individuals in other professions who see that collaborative efforts reap the best outcome. The JCCP will continue to not only bring both clinical and research outcomes to our readers but to encourage us to maintain our course and provide the best chiropractic care we are able to our vulnerable population. Vulnerable because they are young and growing in both body and mind. Our example, our words and our chiropractic care are part of their formative process.

This year, the harvest is in (this was a winter crop! And if I

wasn't mixing metaphors, it would be the catch or the haul, I guess) and being sorted and polished for presentation during the powerful lunar eclipse. Eclipses are known to be "harbingers of change". Look around you, are your patients more stressed and needing your ministrations than you've ever experienced your practice lifetime? Then look at yourself; how are you faring in these times of un-rest and in some cases, un-safety? Whether it's your personal safety in your environment, your safety in choosing your own health care or protecting your privacy, or professionally, legislative changes to your license and scope of practice, information distribution, or even insurance reimbursement?

To be present therapeutically for your patients (many too little to use their worlds to explain their stress level but manifesting it through their body or behaviors instead!) you need to begin with self-care including rest, nutrition and movement (Movement through: Exercise! Breathing! Dancing! Skipping!).

Then, you need to keep learning - talking to each other, reading, attending courses (perhaps we'd consider the greater availability of online education a benefit of this past 3 years?). Restoring your mind and body and building some more plastic connections neurologically are the first order of self-care that can then be extended to our patients not only with what we can contribute but motivating and empowering them to put their own self-care strategies in place because they too (parents) need to take care of themselves so they can take care of the children, but also, we're never too young to learn self-care routines and we have the opportunity to educate the youngest amongst us every time they enter our office with the joy they bring to you at each visit.

The entire world is scrambling to come out of crisis mode and the time is on us to look at transformation rather than continuing to slog through the same mud every day. There are great movers and doers amongst us, and I salute them! Our leaders inspire us and often point us in the most effective direction we can walk. And for many of us, our world is our office and our patients and the ripple effect of what we do for each individual cannot be underestimated, ever, because they too will reach out and touch another and another and another and THAT is how we get the job done!

"E lauhoe mai na wa`a; i ke ka, i ka hoe; i ka hoe, i ke ka; pae aku i ka 'aina."

Everybody paddle the canoes together; bail and paddle, paddle and bail, and the shore will be reached.

Sharon A. Vallone, DC, FICCP
Editor, Journal of Clinical Chiropractic Pediatrics

References:

1. Goertz C, Hurwitz E, Murphy B, Coulter I. Extrapolating Beyond the Data in a Systematic Review of Spinal Manipulation for Non-Musculoskeletal Disorders: A Fall from the Summit. *J Manip Physiol Ther*. 2021.

2. Marson Smith PR WL, Adams C, Chalmers I,. Claims of 'no difference' or 'no effect' in Cochrane and other systematic reviews. *BMJ Evidence-Based Med*. 2019.

3. Hawk C. Comment on Goertz et al article. *J Manipulative Physiol Ther*. 2021;44(6):506-507.

ERRATA

Edits and Correction for Re-publication 3/13/2023

Dorough A. Vallone S. 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

1. RE: Figure 1, pg 1895: "Pop-off: indicates how much (sic) pounds-force….of the fetus". The figure cited for pop-off was 70 pounds of force. The correct value is cited to be between 30-46 pounds force for the vacuum device to pop-off. (29 — Obrien)

2. Incorrect language describing the tethering of oral tissues. The word "fascial" should replace the word ligament on Pg 1897, 2nd column, 3rd paragraph: "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→fascial) structure (20,26,53,54,55,56).

International peer-reviewed chiropractic pediatric history and examination forms for the infant, child, and adolescent

Sue A. Weber BSc, DC, MScAPP (Pediatric musculoskeletal health)
Chairperson/faculty European Academy Chiropractic special interest group in pediatrics (FEAC)
Faculty Royal College of Chiropractors Pediatrics (FRCC)
Practitioner, Stockholm Sweden
dr.sue@telia.com

ABSTRACT

The assessment of the pediatric patient is a specialized area of chiropractic practice, requiring additional knowledge and skills. In recognition of this, European Academy of Chiropractors' special interest group for pediatrics together with an international group of pediatric musculoskeletal experts have expanded on the previously published basic history and examination forms for infants, children and adolescents. The aim of these forms is to further assist the chiropractor in identifying red flags and differentially diagnosing problems in musculoskeletal and mental health as they present throughout growth and development. The process of development of the forms is outlined, and the three forms are presented in this article.

Key Words: chiropractic pediatric, pediatric history form, pediatric examination form, pediatric red flags, pediatric musculoskeletal health.

Introduction

Chiropractic education typically includes a cursory level of education within pediatrics,[1] which varies from institution to institution. Practitioners interested in pediatrics can pursue additional education through continuing professional development courses, a diplomate, a Master's degree or a PhD through a range of providers. Surveys show that the majority of practitioners see children of all ages but feel they have inadequate skills in assessment and treatment.[1]

Triaging musculoskeletal and non-musculoskeletal complaints is of the highest priority for the chiropractor.[2] This is a vital skill and knowledge base for those seeing the pediatric patients to develop, as differential diagnosis and treatment are significantly different than in the adult patient.[3] Children, and particularly infants, are not miniature adults. There are specific and different concerns for each age group which must be addressed with an appropriate history and examination. Basic forms for the infant, child and adolescent have been published to establish a minimum competency for chiropractors who lack advanced education in this area.[4] These forms address skills specifically in musculoskeletal differential diagnosis as well as recognizing for referral the ill child ensuring safe care of this population. Additional exam forms addressing specific pediatric complaints commonly presenting to chiropractors were initiated and intended to complement the basic forms. As these were being drawn up there seemed to be unnecessary repetition so this idea was abandoned. More comprehensive history and exam forms were deemed more appropriate and are presented in this article. They address more age-specific issues in musculoskeletal health, sensory issues, and psychosocial health. For the purposes of safety, the forms include a review of systems, a neurological examination, and red flags. The infant forms include, besides history, a review of the mothers' health and the perinatal period.

AIMS

The European Academy of Chiropractic (EAC) is working to provide cost-free postgraduate education for practitioners. One of the EAC special interest groups (SIG) is pediatrics, where members are working to advance education specific to pediatric practice. The SIG team in pediatrics published in 2021 basic history and exam forms for the infant, child and adolescent.[4] The work since publication of these forms has been to draw up regional forms covering specific conditions within pediatric musculoskeletal health. Due to unnecessary repetition, we decided to enhance and expand the original forms to include these areas along with mental health and wellbeing in the different age groups. Psychosocial health was deemed important as is has become such burden for some children with an increased risk for suicide in adolescents. Pediatric headache history, exam and questionnaire have been recently published separately,[5] so this will be referenced for use in the comprehensive form. An outcome assessment for suboptimal breastfeeding with a musculoskeletal origin has been published recently[6] and is referred to in these forms. Malignancies in children, though rare are different than those presenting in adults.[7] Clinicians should remain abreast of current prevalence statistics and include in the differential diagnosis when appropriate. They are listed in Table 1.

Leukemia
Brain and spinal cord tumors
Neuroblastoma
Wilms tumor
Lymphoma (including both Hodgkin and non-Hodgkin)
Rhabdomyosarcoma
Retinoblastoma
Bone cancer (including osteosarcoma and Ewing sarcoma)

Table 1. Malignancies presenting in childhood

The SIG team engaged the participation of several other chiropractors, most with advanced academic education within pediatrics (Masters or PhD) to review these forms. We intentionally invited practitioners from several different countries to participate to make this an international peer-reviewed pediatric project. A long-term goal is to have these forms implemented internationally to be able to collect data for use in future research projects. The forms are also intended for use as an outline for an academic musculoskeletal pediatric post-graduate education.

The three forms presented with this article are more comprehensive than those published in 2021.[4] They have been designed to organize the pediatric history and examination, giving it consistency, aiding the practitioner in undertaking a thorough assessment. The age groups have been changed to reflect that the period of infancy and toddlerhood were better suited together (0-2 years) than toddler with the young child (Table 2, pages 1961–1968).[6,8,9,10]

The second group is two years to six years, the young child (Table 3, pages 1968–1976)[5,9,10] and the third is 6-16 years which covers the child and the adolescent (Table 4, pages 1976–1983).[5,9,10] The primary focus of the forms is on triaging common MSK and non-MSK presentations in the three pediatric age groups, and on identifying red flags for referral.[12-15] In highlighting non-MSK and red flag presentations, there is an emphasis on safety, particularly identifying and referring the ill child for medical assessment and care.

These forms are intended to address musculoskeletal problems which typically present during the different stages of growth and development. These forms also address more thoroughly issues of mental health, sensory issues, and neurodevelopment in all age groups. They may be helpful not only in reaching the correct diagnosis or diagnoses in order that proper management can be recommended in a timely manner, but that children who need co-management are recognized early in the process.

Process

The pediatric history, examination and red flag forms have been reviewed by 24 different chiropractors, representing 10 different countries: Australia, Belgium, Canada, Cyprus, Denmark, Germany, Malaysia, Norway, South Africa, Sweden, Switzerland, the UK and the United States. The UK was over-represented with nine chiropractors participating. Six participants have a DC degree of which two have a diplomate, fifteen of the participants have MSc in advanced pediatric musculoskeletal health, and three have a PhD with focus in pediatrics. Twenty-three participants work clinically with patients, two of which have graduated less than five years ago.

This process started on December 13, 2022. Thirty-two chiropractors were contacted of which eight chiropractors either did not respond or did not want to participate. One of these chiropractors had a conflict of interest as she is preparing pediatric history and exam forms for financial gain. Emails, tele-conferences, and telephone calls have been made monthly since then to ask for participation, to disperse and discuss each of the forms and to remind participants about the forms. Twenty- three participants who agreed to evaluate the forms reviewed and commented on all three forms, one participant commented on just the child form. Changes were made based on comments and the final iteration was then shared with educators within the field for their final comments. Recent graduates were asked to review the final forms for their completeness and practicality. All participants were asked for their permission to publish their names with educational degrees or diplomate.

Recommendations for chiropractors

It is recommended that chiropractors and other practitioners using manual therapy who are working with pediatrics regardless of their specialty education review these comprehensive forms to enhance their skills. Those without advanced education in pediatrics are encouraged to adopt these more comprehensive forms for clinical practice. Due to the comprehensive nature of the forms, it is understood that the practitioner should focus on the area of complaint and get more detailed information in areas that are problematic. The infant form differs in that it guides the practitioner through a detailed history before the primary complaint. The information gathered gives more clues to the origin of the problem and direction for the exam. Although the forms may be a bit cumbersome for the seasoned practitioner, they guide the newer and less experienced practitioners through a thorough process. These forms address psychosocial health, sensory and neurodevelopmental issues which may be new for the more experienced chiropractor and serve as a good reference in this area. Just as these forms reflect the fact that the child is growing and developing, treatment is also adjusted based on age and development.

A series of recorded lectures to accompany these forms are in progress, discussing key aspects of the history and examination of the pediatric patient. These will be soon available through the European Chiropractic Union home page, European Academy of Chiropractic (EAC) and the EAC Academy.

Conclusion

These forms represent a more comprehensive musculoskeletal assessment of pediatric patients from infancy through adolescence with a goal to ensure safe and effective management. The implementation of these forms should not only raise competence of practitioners, but with widespread use, enable data collection on a large scale for future research. The forms are also to serve as an outline for a postgraduate pediatric education in musculoskeletal health. The work to provide these clinical exam forms is to elevate the safety and quality of care provided by chiropractors to infants, children, and their families.

Limitations

The age groups presented cover more than one area of development. Ideally the pediatric forms should be more specific to distinctive periods of growth and development. Future work in this area would be to further breakdown the age groups and have forms for the neonate, infant, preschooler, child, pre-adolescent, and adolescent.

References:

1. Porcino A, Solomonian L, Zylich S, Gluvic B, Doucet C, Vohra S. Pediatric training and practice of Canadian chiropractic and naturopathic doctors: a 2004-2014 comparative study. *BMC Complementary and Alternative Medicine*. 2017;17(1):512. doi: 10.1186/s12906-017-2024-5.

2. Smith N, Mercer V, Firth J, Jandial S, Kinsey K, Light H, Nye A, Rapley T, Foster HE. Right Path: a model of community-based musculoskeletal care for children. *Rheumatology Advances in Practice*. 2020;4(2). doi: 10.1093/rap/rkaa057.

3. Chance-Larsen K, Backhouse MR, Collier R, Wright C, Gosling S, Harden B, et al. Developing a national musculoskeletal core capabilities framework for first point of contact practitioners, *Rheumatology Advances in Practice*. 2019;3(2)rkz036. doi.org/10.1093/rap/rkz036.

4. Weber S, Miller AS. Chiropractic history and examination forms for the infant, pre-school, and school-aged child. JCCP;2021(20)2:1826-41.

5. Weber S. Pediatric headache questionnaire, exam and history forms for the chiropractor. JCCP;2022(1)1:1871-6.

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

7. Siegel R, Miller K, Wagle, N, Jemal A. Cancer statistics. *CA Cancer J Clin*. 2023; 73:17—48.

8. Matthey S, Barnett B, White T. The Edinburgh Postnatal Depression Scale. *The British Journal of Psychiatry*. 2003;182(4)368-368. doi:10.1192/bjp.182.4.368.

9. Atwell K, Michael W, Dubey J, James S, Martonffy A, Anderson S, et al. Diagnosis and management of hypermobility spectrum disorders in primary care. *The journal of the American board of family medicine*. 2021;34(4)838-848.

10. https://pediatrics.tuftsmedicalcenter.org/The-Survey-of-Wellbeing-of-Young-Children/Age-Specific-Forms

11. Foster HE, Jandial S. pGALS — paediatric Gait Arms Legs and Spine: a simple examination of the musculoskeletal system. *Pediatr Rheumatol*. 2013;11(44). doi.org/10.1186/1546-0096-11-44.

12. Zablotsky B, Black LI, Maenner MJ, Schieve LA, Danielson ML, Bitsko RH, Blumberg SJ, Kogan MD, Boyle CA. Prevalence and Trends of Developmental Disabilities among Children in the US: 2009—2017. *Pediatrics* 2019;144(4):e20190811.

13. Choo YY, Agarwal P, How CH, Yeleswarapu SP. Developmental delay: identification and management at primary care level. *Singapore Med J*. 2019 Mar;60(3):119-123. doi: 10.11622/smedj.2019025. PMID: 30997518; PMCID: PMC6441684.

14. Xue L, Liu C, Xue W, Xue R, Liu P, Wang F. The Role of Nurses in the Management of Respiratory Disorders in Children. *Altern Ther Health Med*. 2022 Jan;28(1):65-71. PMID: 34653020.

15. Frosch M, Mauritz MD, Bielack S, Blödt S, et al. Etiology, Risk Factors, and Diagnosis of Back Pain in Children and Adolescents: Evidence- and Consensus-Based Interdisciplinary Recommendations. Children (Basel). 2022 Feb 2;9(2):192. doi: 10.3390/children9020192. PMID: 35204913; PMCID: PMC8870422.

Pediatric History:

Patient information and consent

Patient name
Patient date of birth
Parent/guardian names, ages, nationality
Legal relationship to child
Family situation
General practitioner/pediatrician
Consent to contact other healthcare practitioners
Consent to exam and care
Date
Consent to use anonymized data confidentially for research

Antenatal health

Perinatal mental health
Maternal Stress
Depression[7]
Maternal prenatal health
Biomechanical status
Mode of conception
Medications, supplements
Migraine headaches
Previous pregnancies

Birth

Duration of pregnancy (gestation)	_____ weeks	_____ days	
Fetal health in pregnancy	_____ 1st stage	_____ hours	
Active fetus	_____ 2nd stage	_____ hours	
Onset of labor	_____ 3rd stage	_____ how long after birth, complications?	
Duration of labor			
Ease of birth			
Fetal presentation			
Fetal distress			
Intervention during labor/birth Emergency C-section			
Medication perinatally analgesia (mother, infant) Antibiotics, drugs, alcohol			
Antibiotics, drugs, alcohol Delivery: hospital, home Injury or stress to neonate after birth (e.g. facial bruising, cephalohaematoma), meconium, jaundice Infant medical conditions at birth Congenital anomalies			

Table 2. Chiropractic pediatric history and exam form ages 0-2 years

Pediatric History, continued:	
Baby's health after birth (APGAR) Delayed cord clamping Postnatal skin to skin Neonatal care Duration NICU Intervention for mother	
Feeding[6]	
How soon after birth breastfed	
Did baby successfully breastfeed directly after delivery	
How/what is baby primarily fed?	
Breast, bottle, other	Type of bottle /nipple
Feeding equally from both sides	
Good latch, efficient feeding	
Concern about tongue-tie 6	
Painful to breastfeed	
Reflux	
Feeding frequency	
Age of introduction solid foods? Interest in food Signs of allergy or intolerance?	
Weight gain and growth Following growth chart	
Any difficulty with or concerns about feeding?	
Crying	
Number of hours crying/day	
Pitch/ intensity	
Timing/ frequency	
Consolability	
Associated symptoms/ behaviors (e.g. pulling ears, scratching eyes)	
Sleeping	
Time and duration (24 hrs)	
Positioning (supine or other)	
Quality of sleep	
Ease of settling, falling and staying asleep Sleeping with mouth open?	
Grinding teeth or night terrors	
Grunting	

Table 2 Continuation. Chiropractic pediatric history and exam form ages 0-2 years

Pediatric History, continued:

Presenting complaint
Parent/guardian concerns Date /mode of onset associated symptoms, timing/course, aggravating and relieving factors
Character, severity, course
Previous episodes
Investigations/outcome
Additional complaints

General health
Diagnosed conditions
Suspected conditions
Medications for mother or baby: Prescribed and over-the-counter What, why, outcome, any side effects
Supplements, D vitamin
Hospital visits, diagnostic testing, imaging When, why, outcome
Other healthcare practitioners seen
Vaccinations — normal schedule followed? Adverse reactions
Family medical history Allergies, migraines, gastrointestinal problems, autoimmune disease

Review of systems
Cardio-respiratory Recurrent coughs, RS virus, wheeze, intervention
Skin Urticaria, eczema, diaper rash
Ears: malformation, hearing Eyes: eye contact, sight Nose: breathing through nose or mouth
Throat
Gastrointestinal Gas, dyschesia, constipation, Poop: consistency and color, Mucous or blood in stool or vomit
Output Number of wet and dirty diapers in 24 hours
Musculoskeletal Positional or postural preference Asymmetry of head or trunk, upright vs. supine

Table 2 Continuation. Chiropractic pediatric history and exam form ages 0-2 years

Pediatric History, continued:

Injuries or falls
Development Does the baby move and interact like other babies the same age? Arms, legs equally active, motor milestones for age?
Any other thoughts or concerns not covered?

Sensory: avoidance or seeking it

Bright light or certain noises
Texture or fit of clothing
Difficulty with brushing hair, teeth or cutting nails

Interoception

Do they know when they become hot, cold, thirsty, hungry, full, tired, excited or ill?
Do they react strongly to certain smells or tastes, will they avoid some foods because of the texture?

Red Flags (indications for referral)

Age group	Sign/symptom
Any age	Labored breathing
	Rib retractions
	Fever: 38°C or over in neonate
	Fewer than 4 heavy wet diapers in 24 hours, signs of dehydration
	Slow or halted growth (weight, length, head circumference)
	Halted or regression of development, loss of skills
	Fractures in non-ambulatory child, unexplained bruising
	From 1 -10 how problematic do you find baby's crying
	Scissored or crossed legs when suspended
Development: 1-3 months	Not responding to loud noises, no eye contact by 6 weeks Not following objects with eyes by 2-3 months
Development: 3-4 months	Not supporting head well Not weightbearing on feet when held upright Persistent flexed positioning of extremity or hand
Development: 4-7 months	Stiff or contracted muscles of extremities, extremity Hypotonia or floppiness of neck or extremities Head not held when pulled from supine to sitting by arms
Development: 8-12 months	Not crawling by 12 months Asymmetry in crawling, e.g. dragging one leg Not standing when supported Not sitting steadily by 10 months

Table 2 Continuation. Chiropractic pediatric history and exam form ages 0-2 years

Pediatric History, continued:

Red Flags (indications for referral)

1 year-old red flags	Constant toe walking
	Lack of reciprocal movement in crawling or walking
	Little awareness of hands, not gesturing with hands
	Not babbling or saying mamma
	Frequent falling or bumping into things

Examination

Observations and vital signs	
General appearance symmetrical movement pattern, tonicity, postural deviations, birthmarks, unusually soft skin, skin color	
Signs of dehydration sunken fontanelle, skin turgor, dry mouth, lack of tearing	
Head, face, eyes, ears, nose Marks, bruising, swelling, discharge, rash, mucous, asymmetry	
Cardiovascular and respiratory Chest wall deformities, respiratory effort, mottling skin, capillary filling	
Abdomen Distention, rigidity, painful behavior	
Vital signs	Weight: _____ kg
	Length: _____ cm
	Head circumference: _____ cm
	Heart rate: _____ bpm
	Respiratory rate: _____ rpm
	Capillary refill: _____ sec
	Temperature: _____ °C

Musculoskeletal examination

Hip screening	
Abduction symmetrical	Clicking in hip with abduction
Supine leg length	Galeazzi sign
Asymmetrical thigh creases or leg positioning	
Tone while suspended	
Hypermobility8	
ROM feet, knees	
Upper limb/shoulder girdle	
Range of motion/tone	
Scarf sign	

Table 2 Continuation. Chiropractic pediatric history and exam form ages 0-2 years

Pediatric History, continued:

Musculoskeletal examination, continued
Signs of asymmetry (biomechanical assessment)
Fixed positioning
Asymmetrical movements
Torticollis: SCM nodule, thickness BL
Plagiocephaly: unilateral occipital flattening, anterior displacement of frontal or maxillary bones
Asymmetrical size of eyes or placement of ears, mouth
Unilateral microcephaly
Infantile scoliosis

Palpation	**Findings**
Static palpation for regional restriction, tension in associated musculature Palpatory hypersensitivity	Cervical: Thoracic: Lumbar: Pelvic: Ribs: Upper extremity: Lower extremity: TMJ: Occiput :
Passive range of motion Joint restriction, muscle tension as indicated	Cervical: Rotation lateral flexion flexion extension: Thoracic: Lumbar: Pelvic: Upper extremity: Lower extremity: Occiput:
Palpation for skull deformity, e.g. open fontanell, cranial synostosis, sutures	
Cranial measurements: Diagonals; Bitemporal; Fronto-occipital	

Neurological examination
Reflexes: Biceps R __ L __; Brachioradialis R __ L __ ; Triceps R __ L __ ; Achilles R __ L __ ; Patellar R __ L __
Sensation: reacts to touch along appropriated dermatome

Primitive Reflexes
Moro reflex (0-4 months)
Rooting (0-4 months)
Palmar grasp (0-4 months)
Galant (0-2/3 months)
ATNR BL (asymmetrical tonic neck reflex) (6 wks)
Babinski (0-walking)
STNR (symmetrical tonic neck reflex)
Parachute 8+ months

Table 2 Continuation. Chiropractic pediatric history and exam form ages 0-2 years

Pediatric History, continued:

Neurological examination, continued

Postural reflexes from 3 months
Head righting bilaterally
Pull to sit without head lag or asymmetry
Supports weight on feet
Ventral suspension
Vertical suspension

Cranial nerve screening	**Normal/abnormal response**
CN II: pupillary light reflex	
CNs III, IV, VI: extra-ocular movements	
CN V: facial sensation, mastication	
CN VII: root response (motor), blink response BL, facial expression	
CN VIII: eyes look to sound	
CN IX, X: suck/swallow/speech	
CN XI: active head rotation	
CN XII: active tongue movement	

Age	**Fine Motor**	**Gross Motor**	**Adaptive**	**Social**	**Communications**
4 weeks		Good head control when held erect	Occasional eye following	Recognizes facial form	Guttural sounds
8 weeks		Head up when prone	Follows	Smiles	Early cooing
12 weeks	Opens hands, grasps all objects, hand-hand, hand-mouth, foot-foot	Balances body weight on lower arms when prone	Regularly looks at objects in hand	Reaches for familiar objects	Laughs
18 weeks		Can shift body weight to one side, lift opp hand to grasp			
6 months	Uses hand in raking motion	Rolling over, both sides	Transfers from hand to hand	Plays with hands	Speech is unclear
9 months	Picks up objects using fingers and thumb	Sits unsupported	Feeds from a cup unassisted	Plays with feet, clearly shows joy / displeasure	Ma-ma, da-da, one or two recognizable words
12 months	Well-developed pincer grip, simultaneously turns 2-3 pages of a book	Crawling established	Holds bottle unassisted	Finger feeds, plays peekaboo	Gestures, jargon
18 months	Turns a page one at a time	Stands unsupported, walks with minimum assistance, runs well, walks upstairs	Builds tower of 2 cubes, feeds self with utensils, scribbles	Understands yes and no, pulls a wheeled toy	4-6 meaningful words, begins two word phrases

Table 2 Continuation. Chiropractic pediatric history and exam form ages 0-2 years

Pediatric History, continued:

Neurodevelopmental[9]
Is your child interested in playing with other children?
Do you have eye contact
When you say a word or wave your hand, will your child try to copy you?
Does your child look if you point to something across the room?
Does your child look at you when you call his or her name?
How does your child usually show you something he or she wants?
Does your child bring things to you to show them to you?

Table 2 Continuation. Chiropractic pediatric history and exam form ages 0-2 years

Pediatric History:
Patient information and consent
Patient name
Patient date of birth
Parent/guardian names
Legal relationship to child
Family situation
General practitioner/pediatrician
Consent to contact other practitioners
Consent to care
Date
Consent to use anonymized data confidentially for research
Primary complaint
Description (ask parent/guardian & child)
Date/mode of onset
Character, course since onset
Possible causes contributing factors
Severity
Injuries or falls
Aggravating factors
Relieving factors
Behavioral changes
Activities affected
Associated symptoms
Previous episodes management
Pre-school attendance, engagement
Teachers or parents' concerns
Additional complaints
Other medical conditions
General health
Diagnosed conditions When diagnosed?
Suspected conditions
Medications
Nutritional supplements

Table 3. Chiropractic pediatric history and exam form for ages 2-6

Pediatric History, continued:
Birth history
Other healthcare professionals consulted
Surgeries, diagnostic testing Interventions: what and why?
Antibiotics
Infections/tick bite
Vaccinations – normal schedule followed? Adverse reactions
Family medical history: allergy, autoimmune, GI, migraine
Review of systems
Respiratory recurrent coughs, mucous, wheeze
Skin urticaria, eczema, rough or discolored patches, bull's eye rash, ring worm, discrete discolored spots (molluscum contagiosum), ecchymosis
Gastrointestinal – pain, vomiting, gas
Output describe odor, color (urine/stool) and texture/form (stool), constipation, dry nights?
Musculoskeletal positional or postural preference asymmetry of head, trunk, or limbs abnormal range of motion head, trunk or limbs
Development moves like other children the same age? symmetry of movement meeting their expected motor milestones?
Nutrition
Diagnosed or suspected allergies/intolerances
Usual diet and any restrictions Feeding behavior
Sleep
Sleep patterns
Ease of falling asleep
Any concerns about sleep
Snoring, night terrors, apnea,
Sleeping with mouth open?

Table 3 continuation. Chiropractic pediatric history and exam form for ages 2-6

Pediatric History, continued:

Activity levels
Physical activity What, how long, how often Total time
Sedentary time
Screen time
Activities

Neurodevelopment
Hyperactivity/ attention
Cognitive/ intellectual development
Emotional and mental health: parental concerns
Behavioral problems
Obsession/compulsion
Tics
Friends
Crisis
Anxiety/depression
Learning issues
Age-appropriate speech and language

Sensory: avoidance or seeking it?
Certain noises or brighter lights?
Bothered by wearing particular clothes
Do they often put non-edible things in their mouth, chew clothes or hair, bite nails?
Are they ok with having their teeth and hair brushed, nails cut

Interoception
Do they know when they become hot, cold, thirsty, hungry, full, tired, excited or ill?
Do they react strongly to certain smells or tastes, will they avoid some foods because of the texture?

Headaches[5] (headache form)	
Headaches	Family history headaches
Migraine headache	
Neck or spinal pain	
Recurrent illnesses	
Fatigue	

Table 3 continuation. Chiropractic pediatric history and exam form for ages 2-6

Pediatric History, continued:
Red Flags (indications for referral)
Sign/symptom
Labored breathing
Rib retractions
Fever
Slow or halted growth Weight, length, head circumference
Halted or regression of development (loss of skills)
Marked difference between left and right sides of body Strength, tone
Marked high or low tone, especially with impact on motor skills/development
Limp
Joint swelling
Is mother worried?
Cyanosis
Disoriented or confused child
Decreased levels of consciousness
Vomiting bile stained (green)
Seizures for the first time/focal seizures
Focal neurological signs
Non-blanching rash with fever
Neck stiffness
Non-weight bearing
Blood in stools or urines
Unexplained bruising
Persistent toe walking

Table 3 continuation. Chiropractic pediatric history and exam form for ages 2-6

Pediatric History, continued:

Developmental milestones

Age		
2 years	Fine motor	Stack 4 blocks, put round or square pegs into holes
	Gross motor	Climbs onto furniture and down, walks in stairs
3 years	Fine motor	Cuts with scissors, string beads, draw a face
	Gross motor	Hops, jumps, rhythmic movements, walks stairs alternating feet
4 years	Fine motor	Draws basic shapes, draws a house, uses pencil with good control
	Gross motor	Climbs ladders, navigates obstacles when running
5 years	Fine motor	Copies letters and numbers, ties shoelaces
	Gross motor	Walks narrow line, plays ball games, skips on alternate feet
6 years	Fine motor	Can write their names, dress themselves
	Gross motor	Good balance, run, jump, skip easily

Skills from 3 years
Jump in place with both feet
Climb up and go down a toddler slide
Pedal tricycle
Walk up and down stairs when one hand being held by an adult
Stand briefly on one foot when one hand being held by an adult
Walk backwards

Skills from 4-5 years
Hop on one foot
Tandem gait
Toe to heel gait
Do a somersault/ hop
Walk up and down stairs without help
Walk forward and backward easily
Stand on one foot > 9 seconds

Table 3 continuation. Chiropractic pediatric history and exam form for ages 2-6

Pediatric History, continued:

Examination

Observations and vital signs

General appearance: movement pattern, handedness, postural deviations, torticollis, plagiocephaly, leg length, asymmetry
Skin, joints[8] soft, extensible skin, obvious signs of hypermobility, birthmarks
Head, face, eyes, ears, nose marks, bruising, swelling, discharge, rash, mucous, asymmetry
Cardiovascular and respiratory chest wall deformities, respiratory effort, color
Abdomen distention, rigidity, umbilicus
Social interaction child and parents, child and practitioner
Measurements

Weight: _____ kg/lbs

Length: _____ cm/in

Heart rate: _____ bpm

Respiratory rate: _____ rpm

Temperature: _____ °C

Overweight/ obesity: _____

Neurological examination

Cranial nerve screening
CN II: Pupillary light reflex
CNs III, IV, VI: extra-ocular movements
CN V: facial sensation, tone mastication
CN VII: symmetrical blink, Facial expression
CN VIII: eyes look to sound
CN IX, X: speech, swallow
CN XI: active head rotation
CN XII: active tongue movement
Eye exam H
Consensual eye movement
Gower sign
Heel/toe walk
Rapid alternative movements
Romberg: standing balance test eyes closed
Conjugated eye tracking
Finger to nose
Persistent primitive reflexes

Table 3 continuation. Chiropractic pediatric history and exam form for ages 2-6

Pediatric History, continued:

Examination, continued

Muscle stretch reflexes Present/absent/atypical	R	L
Biceps		
Brachioradialis		
Triceps		
Patella		
Hamstring		
Achilles		
Babinski		
Clonus		

Orthopedic/ Musculoskeletal examination

Palpation	Findings
Active and passive range of motion Spine, extremities as indicated	Cervical: _____ Occiput: _____ Thoracic: _____ Costovertebral/costosternal: _____ Lumbar: _____ Pelvic: _____ Upper extremity: _____ Lower extremity: _____
Static and motion palpation for regional restriction, tenderness Spine, extremities — as indicated Paraspinal symmetry/hypermobility	Occiput: _____ Cervical: _____ Thoracic: _____ Lumbar: _____ Pelvic: _____ Upper extremity: _____ Lower extremity: _____
Temporomandibular joints	
Hip: alignment/ function	
Knees: alignment/ function	
Feet/ankle: alignment/function	
Shoulder girdle: alignment/ function	
Adams test	
Leg length	
Scoliosis	
Hyper/hypo mobility	
Posture	
Muscle tonus/ symmetry/ strength	
Gait/ limp	
Leg pain: intermittent evening	

Table 3 continuation. Chiropractic pediatric history and exam form for ages 2-6

Pediatric History, continued:

Examination, continued

PGALs Check[10]

Gait — normal heel toe
'Touch the sky'
'Touch your toes'
'Hands out'
'Fists/turn'
'Fingers touch'
'Jaw range of motion'

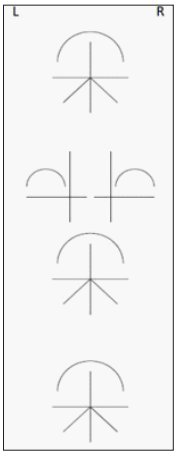


Table 3 continuation. Chiropractic pediatric history and exam form for ages 2-6

Pediatric History:

Patient information and consent

Patient name		
Patient date of birth		
Parent/guardian names, nationalities		
Legal relationship to child		
Family situation		
General practitioner/pediatrician		
Consent to contact other practitioners		
Consent to care		
Date		
Consent to use anonymized data confidentially for research		

Primary complaint

Description (ask parent/guardian & child)		
Location of pain		
Date/mode of onset		
Course since onset		
Duration	intensity	frequency
Character/ severity of pain		
Antalgia	radiculopathy	
Possible causes/contributing factors		
History physical trauma, fall, injury		
Aggravating factors		
Relieving factors		
Associated symptoms		
Night pain	relief with medication	
Pain with movements		
Pain on impact		
Pain when sitting		
Pain changing positions		
Interference with activities		
Behavioral changes		
Previous episodes/ management		
Activities after school		
hours training	hours high impact	frequency
Competitive Sports		
Sedentary Time	hrs screens use	
Ergonomics school	home	
Family history scoliosis	bracing	

Table 4. Pediatric musculoskeletal history and exam form for ages 6-16 years

Pediatric History, continued:

Primary complaint, continued

Hx asymmetry	
Hip dysplasia	torticollis
Toe walking	
Coordination/motor skills	
Secondary or other complaints	
Sleep issues: (quality, snoring, apnea, night terrors)	
Headache (HA) headache questionnaire[5]	Family history HA/migraine
Vision	

Other medical conditions

General health
Diagnosed conditions When diagnosed? Treatment, outcome
Suspected conditions
Medications Prescribed and other
Nutritional supplements
Hospital visits/admissions
Other healthcare professionals consulted
Surgeries
Radiological history, diagnostic tests
Infections, tick bites
Vaccinations — normal schedule followed? adverse reactions
Family medical history rheumatological dz, hip disorders, allergy, autoimmune, genetic conditions, gastrointestinal, connective tissue disorder
Smoking, alcohol, drugs

Nutrition

Diagnosed or suspected allergies/intolerances
Selective diet and any restrictions

Meals per day: _____

Snacks per day: _____

Number of sugar portions/day: _____

Does child eat breakfast: _____

Table 4 continuation. Pediatric musculoskeletal history and exam form for ages 6-16 years

Pediatric History, continued:

Review of systems

Respiratory: asthma recurrent cough, mucous, wheeze		
Skin: urticaria, eczema, ecchymosis, bull's eye rash, rough or discolored patches, acne		
Sign of connective tissue disorder[8] poor posture, hypermobility Pes planus		
Gastrointestinal pain, vomiting, gas		
Output: Primary or secondary enuresis, constipation, loose stools		
Age of menarche	discomfort	
Psychological		
Any history or signs of emotional trauma or abuse		bullying
Social skills		well-being
Behavioral issues	anxiety	Hyperactivity
Conduct		issue with friends or family
Bereavement	attention	concentration
Depression	harmful self behavior	
School attendance, engagement		
Sensory issues: texture, taste, smell food		
Texture clothing	issues with light or noise	
Neurodivergence	gender issues	
Involuntary movements, tics		
Obsession: gets stuck in a particular thought process		
Compulsion: repeatedly have the same concern or anxiety		
Does not show empathy		
Has trouble sitting still for a long period of time		
Finds it hard to make friends		
Unusual fears or worries		
Extreme rigidity about routines		

Table 4 continuation. Pediatric musculoskeletal history and exam form for ages 6-16 years

Pediatric History, continued:
Red Flags (indications for referral)
Labored breathing
L thoracic curve
Slow or halted growth Growth curve/chart
Halted or regression of development (loss of skills)
Marked difference in strength between L/ R sides of body
Marked high or low tone, especially impacting motor skills/development
Persistent toe-walking
Swelling of a joint for longer than 6 weeks
Recent trauma with suspected fracture
Recent infection with suspected ongoing infection
Signs and symptoms of diabetes type I
Systemic upset (malaise, weight loss, night sweats)
Lymphadenopathy
Unremitting bone pain
Incongruence between history and physical examination
Inability to weight bear
Persistent pain and morning stiffness of more than 30-60 minutes
Pain waking the child at night
Can not hop, skip or jump
Can not dress independently

Table 4 continuation. Pediatric musculoskeletal history and exam form for ages 6-16 years

Pediatric History, continued:

Examination

Observations and vital signs

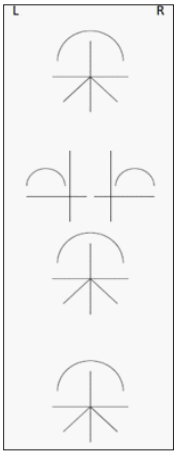
General observations appearance, movement pattern, coordination, skin, handedness		
Skin Beighton 4/9, narrow palate, general or local joint laxity, soft extensible skin		
Head, face, eyes, ears, nose, throat marks, bruising, swelling, discharge, rash, mucous, asymmetry		
Eye tracking Eye Exam 'H'		
Cardiovascular and respiratory chest wall deformities, respiratory effort, cyanosis		
Musculoskeletal positional or postural preference asymmetry of head, trunk, or limbs abnormal ROM of head, trunk or limbs		
Development for age does the child move and interact like other children the same age?	Weight: _____ kg/lbs Length: _____ cm/in Heart rate: _____ bpm Respiratory rate: _____ rpm Temperature: _____ °C Overweight/ obesity: _____	

Posture, symmetry including extremity positioning

Scoliosis	Adams test	Kyphosis	Lordosi
Asymmetry: thoracic curve/ lumbar curve/ double curve/rib hump			
Forward head posture	Torticollis	Leg length	
Standing	Sitting	Lying	
Symmetry: ilia, jaw, ribs			
Feet	Knees	Hips	
Gait	contralateral pattern during walking		
pGALS10: Functional assessment			
Heel walk	toe walk		
Spread fingers	supinate hands and make fist		
Pinch thumb to fingertops	joint swelling		
Put backs of hands together	squeeze metacarpals		
Put hands behind neck, elbows out			
Active flexion/extension of knees	internal rotation hips		
Open mouth wide and put 3 of their fingers in mouth			
Cervical lateral flexion	lateral flexion lumbars		
Forward flexion head	reach for sky, look up		

Table 4 continuation. Pediatric musculoskeletal history and exam form for ages 6-16 years

Orthopedic examination

Palpation : skin, muscles and joints	Findings
Active and passive range of motion symmetry as indicated	Cervical: _____ Occiput: _____ Thoracic: _____ Lumbar: _____ Pelvic: _____ Upper extremity: _____ Lower extremity: _____
Static and motion palpation for regional restriction, tenderness range of motion as indicated  	Cervical: _____ Occiput: _____ Thoracic: _____ Lumbar: _____ Pelvic: _____ Upper extremity: _____ Lower extremity: _____

Regional exam: Attention to size, bulk and tone of muscles

	Normal/abnormal findings
Hip	
Shoulder	
Knee	
Feet/ankle	
TMJ	
Hypersensitivity to pain	
Valsalva	
SLR	

Table 4 continuation. Pediatric musculoskeletal history and exam form for ages 6-16 years

Neurological examination	
Walk heel-toe with good balance	
Stand steadily with feet together, eyes closed	
Stand steadily on one leg, eyes open	
Stand steadily on one leg with eyes closed	
Heel-toe walk	
Finger-to-nose	
Rapid alternating movements	
Cranial nerve screening	**Normal/abnormal response**
CN I CN II: Pupillary light reflex	
CNs III, IV, VI: extra-ocular movements	
CN V: facial sensation, masseter/temporalis motor	
CN VII: blink response, facial expression	
CN VIII: hearing screen	
CN IX, X: speech, swallowing	
CN XI: active head rotation	
CN XII: active tongue movement	
Muscle stretch reflexes	**Normal/abnormal response R / L**
Biceps	
Brachioradialis	
Triceps	
Patella	
Hamstring	
Achilles	
Babinski flexor response	
Sensation	
Strength	
Clonus	
Retained primitive reflexes	
Persistent toe walking	

Table 4 continuation. Pediatric musculoskeletal history and exam form for ages 6-16 years

Chiropractic management of non-synostotic deformational plagiocephaly in the Netherlands: a cross-sectional study

Camille Verfaillie, MChiro, MSc[1], Alister Du Rose, PhD, PGCert, MChiro[2], Amy Miller, MSc, PhD, FEAC (Paeds)[3]

[1]Chiropractor, Izegem, Belgium
[2]Senior Lecturer (Clinical Sciences), AECC UC, Bournemouth, UK
[3]Lecturer (Clinical Sciences), AECC UC, Bournemouth, UK
Corresponding Author, Camille Verfaillie; Email: camilleverf@hotmail.com

ABSTRACT

Background: Non-synostotic deformational plagiocephaly (NDP) is a common condition affecting 48% of infants. It is frequently cited as a reason for presentation to a chiropractor, however little is currently known about chiropractic management of NDP. This cross-sectional study aims to begin to address this literature gap. **Objectives:** 1) To determine the characteristics of chiropractic management of non-synostotic deformational plagiocephaly (NDP) in the Netherlands and; 2) To investigate the type and number of treatments chiropractors expected for full resolution of the head turning preference. **Methods:** Cross-sectional survey of registered chiropractors in the Netherlands. **Results:** Seventy-eight chiropractors completed the survey, of which 86% (N=67) treated pediatric patients, and of which 73% (N=38) treated infants with NDP. The most common presentation was head turning preference (with or without NDP) (75%, N=39) for 0-11-month-olds and the most common treatment technique was 'touch and hold' (65%, N=33). Participating chiropractors reported 'no side effects' (39%, n=20) more commonly than any specific side effects. On average, participating chiropractors expected 4 treatments for full resolution of the head turning preference. **Conclusion:** Chiropractors in the Netherlands who treat children frequently manage cases of NDP. In line with current evidence, participating chiropractors often attributed NDP to a head turning preference. There is currently limited clinical evidence on the effectiveness of management techniques for head turning preference and NDP, this should be the focus of future research.

Key Words: Non-synostotic deformational plagiocephaly, plagiocephaly, head turning preference, chiropractic, the Netherlands.

Introduction

Chiropractic is:

> "a health profession concerned with diagnosis, treatment and prevention of mechanical disorders of the musculoskeletal system, and the effects of these disorders on the function of the nervous system and general health"[1] and is categorised as complementary and alternative medicine (CAM).[2,3]

Approximately 5-17% of global chiropractic practice are represented by pediatric patients.[4] A recent international demographic chiropractic study, based on 1,498 respondents from 17 countries over 6 continents, showed that 90% of chiropractors accepted pediatric patients.[5] Despite its widespread use, there is debate over the appropriateness of pediatric chiropractic care, with concerns relating to safety, effectiveness, presentations/complaints[6] and a paucity of high-quality research.[4,6] In terms of safety, studies have shown that mild side effects, which are self-limiting, following pediatric chiropractic care comprise the majority of side effects with an incidence of 1% in patients under three.[7] Systematic reviews demonstrate no deaths reported, and that in the rare cases of serious adverse events (requiring hospitalization), underlying pre-existing pathology preceded.[8,9]

Controversy around chiropractic care for infants is also driven by the variety of musculoskeletal and non-musculoskeletal complaints which are treated in chiropractic practices.[10] Research claims about beneficial effects of chiropractic on commonly treated non-musculoskeletal conditions include sleep issues, asthma, otitis media, and even jet lag.[11] However, evidence supporting treatment of non-musculoskeletal conditions are typically of low scientific value, consisting of clinical experience and case studies.[11] Most pediatric patients are presented for chiropractic care with musculoskeletal problems, the frequency of which increases with age, from 23-33% in preschool children and 75-84% in teenagers.[12,13]

One of the most common orthopedic conditions in infants is non-synostotic deformational plagiocephaly (NDP), with prevalence estimates of 48% of infants.[14] NDP is defined as cranial asymmetry manifesting in flattening

of the skull secondary to external forces without fusion of the skull sutures (synostosis).[15,16] The incidence of NDP has increased five-fold since the 'Back to Sleep' campaign and the American Academy of Pedatrics' recommendation for healthy new-borns to sleep supine to reduce the risk of sudden infant death syndrome.[17-19] NDP is associated with cosmetic consequences and has been associated with neurodevelopmental delay, although no causal relationship has been demonstrated.[20-22] There is some evidence that infants with NDP but no neurodevelopmental delay may later develop delay in childhood, resulting in language disorders, attention deficits and learning disabilities.[23] While evidence around NDP and developmental delay is still emerging, it could be argued that the cosmetic element alone is worth preventing.

According to a cross-sectional study by Roby et al. (2012)[24] 38% of infants with NDP and/or brachycephaly had abnormal facial characteristics and have a 2% chance of having those facial deformities persist into adolescence when left untreated. Two other studies in preschool-aged children demonstrated a prevalence of remaining deformity of 3.3% at two years of age[26,27] and 2.4%-4% at three years of age.[26,28] Robinson and Proctor (2009) estimate that 0.5-1% of children will show obvious cranial deformities when entering school.[25] Hence, in a small proportion of infants with NDP, facial and cranial deformities persist into childhood. The craniofacial deformity and possibly consequential teasing, bullying or embarrassment is one of the most reported parental concerns relating to their child's NDP[29] which might be valid because it has been shown that facial "attractiveness" significantly influences the behaviors of caregivers,[30] social interactions with peers[31-33] and teachers' expectations about intelligence and popularity of the child.[34] Two very recent studies also showed that persons with craniofacial deformities are susceptible to (cyber)bullying[35] and are at higher risk of psychosocial problems.[36]

Sleeping supine with head preference predisposes to NDP.[36-38] This is why head positional preference is discussed in this study. There is currently limited research exploring pediatric chiropractic in the Netherlands. Whilst four studies have been conducted[39-41] none have investigated the clinical characteristics of pediatric chiropractic care and the treatment of NDP in infants.

There are many different chiropractic treatment techniques used with pediatric patients, and chiropractors adapt force and speed used in manual therapy to match the child's age and development.[42] Due to the wide range of treatment techniques, this study investigates association between treatment techniques and the total number of treatments expected for full resolution of the head turning preference, as well as determining the characteristics of how chiropractors manage NDP, all providing new insight into chiropractic management of NDP in the Netherlands.

Given that NDP and head turning preference are reported by parents as reasons for presenting their infant to the chiropractor, and the limited evidence for chiropractic management of NDP, this paper sets out to describe chiropractors' experiences and perceptions of this common problem and may serve as a starting point for future research into this condition. This paper does not provide evidence of effectiveness but does highlight the frequency of the condition, treatment types and side effects.

Methods

The research design was a cross-sectional study of practicing chiropractors in the Netherlands. Ethical approval (E142/03/2021) was obtained from AECC University College and Nederlandse Chiropractoren Associatie's Science Committee.

Sample

The inclusion criteria were practicing chiropractors in the Netherlands who were registered with one of the Dutch chiropractic associations: Dutch Chiropractic Federation (DCF), Christelijke Chiropractoren Associatie (CCA) or Nederlandse Chiropractoren Associatie (NCA). Participating chiropractors also had to comprehend written English.

According to Fincham (2008), the response rate should approach 60% to enable appropriate generalization.[43] In previous cross-sectional surveys about pediatric chiropractic care, Lee et al. (2000) achieved a response rate of 60% (90 respondents),[44] and Durant et al. (2001) achieved a response rate of 57% (77 respondents).[45] A previous demographic survey study in the Netherlands about chiropractic achieved a response rate of 78% (94 respondents).[41] Based on these previous similar studies and the scientific quality provided by response rates of >60%, this was the recruitment goal for this study (60%, n=296).

Data collection

Data were collected via a one-time online anonymous questionnaire. The questionnaire was hosted on Jisc Online Surveys.

Face and content validity of the questionnaire was established using a panel of three experts: a Lecturer in Research Methods at AECC UC, a Dutch chiropractor with knowledge of advanced research methods, and the Course Lead for the MSc APP Pediatric Musculoskeletal Health at AECC UC. These experts were asked for feedback and minor modifications were made based on this, including wording, content, and English language.

This study is part of a larger study. The overall questionnaire

concerned more general information about pediatric chiropractic management, but data specifically related to infants and NDP was pulled from that questionnaire and used for this study.

The information sheet, instructions, and survey were emailed to the Dutch chiropractic associations for distribution to their members. All associations agreed to participate. A reminder email was sent to association members after ten days, the survey was closed one week after this. Timelines were limited as this was a MSc project.

Data analysis
Data were transferred into Excel and IBM SPSS Statistics 24 for analysis. Descriptive statistics were used to quantify the demographic profile of participating chiropractors. A one-way ANOVA test was performed to determine any association between treatment techniques used and number of treatments needed for full resolution of the head turning preference (Table 1).

Results
Seventy-eight responses were received from a total of 493 members of the chiropractic associations, a 16% response rate, significantly less than the 60% target. Of the 78 responses, 86% (N=67) treated pediatric patients. The results presented are all based on the chiropractors' report, rather than medical records.

	Sum of Squares	Df	Mean Square	F	Sig.
Between Groups	88.702	43	2.063	2.06	0.30
Within Groups	3	3	1	3	5
Total	91.702	46			

Table 1. One-way ANOVA - association between treatment techniques used and number of treatments needed for NDP.

Presentations
Participating chiropractors could choose more than one answer. For the age group 0-11 months, the most common presentation reported was head turning preference (with or without NDP) (75%, N=39) and most participants (73%, N=38) treated NDP.

Treatment techniques
Participating chiropractors could choose more than one answer. The definitions of the treatment techniques can be found in Table 2. The most common treatment technique for NDP was 'touch and hold' (68%, N=26), followed by cranial techniques (58%, N=22) and exercises and advice to parents both at 55% (N=21) (Table 3). The data do not specify where the touch and hold technique was applied.

Referral and co-management patterns
Participating chiropractors could choose more than one answer. Infants with NDP were most commonly referred to participating chiropractors by midwives (55%, N=22) and physiotherapists (48%, N=19) (Table 4, page 1988). In terms of outward referrals, participating chiropractors most frequently referred infants with NDP to GPs (60%, N= 24) and physiotherapists (50%, N=20), either for co-management or sole management by that practitioner. The inward and outward referrals were two different questions and do not necessarily relate to the same patients.

Age and number of treatments
The mean age at which NDP was mostly encountered was 1.53 months (M=1.53, SD=0.554) (Tables 5 and 6, page 1988). If NDP was associated with a head turning preference, participating chiropractors expected four treatments on average (M=4.15, SD 1.562) for full resolution of the head

Activator technique	A handheld, spring-loaded instrument that provides a specific low-force-type thrust to restricted joints of the spine or extremities
Applied Kinesiology (AK)	Correction of muscle weakness/imbalance via the nervous system, lymphatic system, vascular system, and nutrition
Diversified technique	SMT (spinal manipulative therapy) including a high-velocity, low-amplitude (HVLA) thrust to areas of restricted joints of the spine or extremities
Neural Organization Technique (NOT)	Technique to reorganize and enhance the autonomic nervous system's function by using combinations of specific SMT, SOT, AK, and acupressure (neuro-lymphatic and neuro-vascular points) with coordinated breathing to improve the function of the systems controlled by the autonomic nervous system
NeuroImpulse Protocol (NIP)	A very gentle technique with focus on neurological precision. Not intended to restore joint function but to restore normal neurological function and repositioning the body through the central nervous system
Sacro-occipital technique (SOT)	A technique that focuses on the relationship between the sacrum and occiput
Steve Williams' technique	Technique that focuses on functional pediatrics, SOT and craniopathy
Toggle recoil	Fast but light thrust, quickly removing chiropractor's hands away from the contact point
Touch and hold	A gentle pressure is applied to the fixated structure until the practitioner feels the tension releasing

Table 2. Definition of techniques used by participating chiropractors.

Treatment techniques	Responses N	% of cases
Activator technique	14	7%
Applied kinesiology	2	5%
Cranial techniques	22	58%
Diversified technique	3	8%
Exercises	21	55%
Advice to parents	21	55%
NeuroImpulse Protocol	6	16%
Sacro Occipital Technique	12	32%
Soft tissue work	16	42%
Steve Williams' technique	17	45%
Toggle recoil	3	8%
Touch and hold	26	69%
Neural Org. Technique	1	3%
Total	164	432%

Table 3. Treatment techniques for NDP.

turning preference (Table 6, page 1989). There was no statistically significant relationship between treatment techniques used for NDP and number of treatments estimated for full resolution of the head turning preference, determined by the one-way ANOVA (p = .305) (Table 1).

Discussion

According to Hestbaek & Stochkhendahl (2010),[46] musculoskeletal conditions are the most common presentations/diagnoses in children which was also shown in this study. This is consistent with Durant et al. (2001),[45] Verhoef and Papadopoulos (1999),[47] Hestbaek et al. (2009)[48] and Miller (2010).[49] This might be explained because musculoskeletal complaints frequently present in general pediatric practice as well,[50] and chiropractors are known to be musculoskeletal specialists.[48]

Pediatric chiropractic practice in the Netherlands is common with 86% (N=67) of participating chiropractors treating patients under 18. It is not known why the other chiropractors did not respond, however given the high proportion of respondents who treated pediatric patients, it may be that these chiropractors were more inclined to respond to the survey.

Although NDP was the least commonly chosen presentation for the 0-11-month-olds, the most common presentation was head turning preference (with or without NDP) (Table 7, page 1990) and the majority of participating chiropractors indicated in the survey that they treated NDP.

The mean age at which NDP was encountered was 1.53 months (M=1.53, SD=0.554) (Table 6, page 1989)

Professionals	Responses N (percent of cases)	
	Professionals to which chiropractors refer infants	Professionals that refer infants to chiropractors
Another chiropractor	9 (23%)	11 (28%)
Craniosacral therapist	1 (3%)	1 (3%)
General practitioner	24 (60%)	9 (23%)
Homeopath	2 (5%)	2 (5%)
Lactation consultant	-	13 (33%)
Maternity care physician	-	13 (33%)
Midwife	-	22 (55%)
Physiotherapist	20 (50%)	19 (48%)
Specialist at the hospital	12 (30%)	2 (5%)
(Specialist at) tongue tie clinic	-	1 (3%)
Doula	-	1 (3%)
Osteopath	7 (18%)	-
None	4 (10%)	6 (15%)
Total	79 (198%)	100 (250%)

Table 4. Referral patterns for NDP.

which aligns with existing prevalence data demonstrating increases in NDP in healthy infants up to 16 weeks of age.[40]

If NDP was associated with a head turning preference, participating chiropractors expected four treatments on average (M=4.15, SD 1.562) for full resolution of the head turning preference (Table 6). This represents a relatively rapid resolution compared to eight chiropractic treatments reported by Hash (2014),[14] and three to four months of chiropractic care recommended by Davies (2002).[51] However, it is important to note that Hash (2014) and Davies (2002) described these timelines for full resolution of NDP, rather than head turning preference alone.[14,46] This estimate of four treatments was consistent with Saedt et al. (2018), where the head turning preference resolved in averagely 3.5 treatments of manual therapy with the greatest effects obtained after 1.8 treatments.[40]

Participating chiropractors reported a relatively young infant population and relatively low numbers of treatments for resolution of the head turning preference and it may be plausible that older infants may require additional/longer term treatment, although there is no definitive evidence to confirm at this stage and differences in resolution times across different ages should be addressed in future research.

In our study, there was no statistically significant relationship between treatment techniques used for NDP and number of treatments needed for full resolution of the head turning preference, indicating that effects are specific to the individual. Participating chiropractors of this study indicated 'touch and hold' and cranial techniques as the two most common treatment techniques for NDP, followed by exercises and advice to parents as the third most common treatment types. Likewise, cranial techniques, including decompression of the occiput, frontal bone lift and traction of the temporals, were also a large component of the treatment plan in the study of Hash (2014).[14]

Hash (2014), Davies (2002) and Cabrera-Martos et al. (2016)

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	0-3 months	20	38.5	50	50
	3-6 months	19	36.5	47.5	97.5
	6-9 months	1	1.9	2.5	100
	Total	40	76.9	100	
	System	12	23.1		
Missing Total		52	100		

Table 5. Ages at which the chiropractors encounter NDP.

		At what age does the chiropractor encounter NDP the most?	If plagiocephaly is caused by a head turning preference, how many treatments does the chiropractor usually expect, on average, for full resolution of the head turning preference?
N	**Valid**	40	40
	Missing	12	12
Mean		1.53	4.15
Median		1.5	4
Mode		1	4
Std. Deviation		0.554	1.562
Variance		0.307	2.438
Percentiles	25	1	3
	50	1.5	4
	75	2	5

Table 6. Statistics on age and number of treatments for NDP.

Presentations	Responses N (percent of cases)
Check up without complaints	29 (56%)
Colic	32 (62%)
Feeding difficulty	19 (37%)
Head turning preference (with or without (non-synostotic) plagiocephaly)	39 (75%)
Motor development delay/issues	9 (17%)
Musculoskeletal conditions	12 (23%)
Scoliosis	4 (8%)
Sleeping difficulty	30 (58%)
Frequent colds	1 (2%)
Non-synostotic deformational plagiocephaly	1 (2%)
Ear infection	1 (2%)
Tongue tie	1 (2%)
Birth trauma	1 (2%)
Headache	-
Nocturnal enuresis	-
Neurodevelopmental issues	-
Emotional control	-
Brain injury	-
Gait abnormality	-
Abdominal issues including pain and/or constipation	-
Swimming issues	-
Extremity joint pain	-
Low back pain	-
Neck pain	-
Nocturnal enuresis	-
Performance improvement	-
Sports injury	-
Whiplash Associated Disorder	-
Neurological disorder	-
Learning disorder	-
Not applicable for participant	8 (15%)
Total	**187 (360%)**

Table 7. Presentations 0-11 months of age.

reported benefit of chiropractic/manual therapy for the management of NDP without side effects.[14,17,46] The study of Saedt et al. (2018) is an observational study without randomization and control groups so no conclusion can be made about the effectiveness of upper cervical manual therapy. Nevertheless, NDP appeared to improve with upper cervical mobilization techniques. This is believed to be beneficial as NDP is often caused by upper cervical dysfunction resulting in actively and passively restricted cervical ROM.[40] Fludder and Keil (2020) found restricted passive cervical ROM in 92% of children with NDP. They also showed 79% of children under the age of one suffered from restricted passive cervical ROM, of which 60% also showed indication of NDP.[42] These factors might explain why manual therapy can be of benefit in management of NDP.

Limitations

This study comes with limitations. Firstly, there is non-response bias. The survey was voluntary and resulted in a relatively low response rate of 16%, significantly below the

target of 60%. This means the results only reflect the practice of a small proportion of chiropractors in the Netherlands and limits generalizability. There is no data to explain why participation was low. It can be hypothesized that it may be attributed to the short time frame to complete the survey and the timing. At the time of distribution, chiropractic clinics only had been officially open for a few weeks since the second COVID-19 lockdown as chiropractic is not considered an essential service in the Netherlands.

Chiropractors may well have had other priorities at this time, and this may further account for the low response rate. Future studies could use multiple means of promoting the study such as social media and not email alone. However, the sample size is equivalent to previously published studies in this area. It needs to be considered that this data was pulled from a larger study. The information provided was dependent on the respondent's recollection and accuracy, potentially leading to recall bias. Participating chiropractors with a particular interest in pediatrics might have been more likely to participate which may have introduced selection bias, chiropractors who infrequently treat pediatric patients may be underrepresented in this study and there may be an unreported difference in their practice and management of NDP.

Future studies may want to address specific age groups to get more detailed results, as well as add a longer time frame for response. This is however the first study to investigate pediatric chiropractic practice in the Netherlands. Further research is needed to address this paucity in quality and quantity of data. Initially, a prospective study of infants undergoing chiropractic management, using valid parent-reported outcomes about head turning preference and objective measures such as measuring the distance from ear to external occipital protuberance with pre- and post-treatment data collection to assess for change. Future research may also want to address cost/benefit ratio and rates of satisfaction.

Conclusion

This study was based on data collected for a larger study, which will be published separately. Most participating chiropractors treated pediatric patients, mainly using the low-force technique 'touch and hold'. The most common presentation was head turning preference (with or without non-synostotic plagiocephaly). Non-synostotic plagiocephaly is typically encountered at around 1.53 months and treated with 'touch and hold' and cranial techniques. If it was associated with a head turning preference, four treatments were expected on average to achieve full resolution of the head turning preference. Participating chiropractors reported 'no side effects' more commonly than any specific side effects.

This study serves to raise awareness of the high occurrence of non-synostotic deformational plagiocephaly, and the role chiropractors can play in its management, which is in line with current recommendations with regards to treatment types, home advice and number of treatments.

References:

1. wfc.org [Internet]. Toronto: World Federation of Chiropractic [cited 2021 October 3]. Definition of chiropractic. Available from: https://www.wfc.org/website/index.php?option=com_content&view=article&id=90.

2. Alcantara J, Ohm J, Kunz D. The safety and effectiveness of pediatric chiropractic: A survey of chiropractors and parents in a practice-based Research Network. *EXPLORE*. 2009;5(5):290—5.

3. Gleberzon B, Arts J, Mei A, McManus E. The use of spinal manipulative therapy for pediatric health conditions: a systematic review of the literature. *J Can Chiropr Assoc*. 2012;56(2):128-41.

4. Marchand AM. Chiropractic care of children from birth to adolescence and classification of reported conditions: an internet cross-sectional survey of 956 European chiropractors. *J Manipulative Physiol Ther.* 2012;35(5):372—80.

5. Doyle MF, Miller JE. Demographic profile of chiropractors who treat children: a multinational survey. *J Manipulative Physiol Ther*. 2019;42(1):1—11.

6. Spigelblatt L. Chiropractic care for children: Controversies and issues. *J Paediatr Child Health*. 2002;7(2):85-9.

7. Miller JE, Benfield K. Adverse effects of spinal manipulative therapy in children younger than 3 years: a retrospective study in a chiropractic teaching clinic. *J Manipulative Physiol Ther*. 2008;31(6):419—23.

8. Vohra S, Johnston BC, Cramer K, Humphreys K. Adverse events associated with pediatric spinal manipulation: a systematic review. *PEDIATRICS*. 2007;119(1).

9. 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. *J Manipulative Physiol Ther*. 2015;38(9):699—712.

10. Allen-Unhammer A, Wilson FJ, Hestbaek L. Children and adolescents presenting to chiropractors in Norway: national health insurance data and a detailed survey. *Chiropr Man Ther*. 2016;24(1).

11. Gotlib A, Rupert R. Chiropractic manipulation in pediatric health conditions — an updated systematic review. *Chiropr Osteopat*. 2008;16(1).

12. Miller J. Demographic survey of pediatric patients presenting to a chiropractic teaching clinic. *Chiropr Osteopat*. 2010;18(1).

13. Hestbaek L, Jørgensen A, Hartvigsen J. A description of children and adolescents in Danish chiropractic practice: results from a nationwide survey. *J Manipulative Physiol Ther*. 2009;32(8):607—15.

14. Hash J. Deformational plagiocephaly and chiropractic care: A narrative review and case report. *J Clin Chiropr Pediatr*. 2014;14(2):1131-8.

15. Collett B, Gray K, Starr J, Heike C, Cunningham M, Speltz M. Development at age 36 months in children with deformational plagiocephaly. *PEDIATRICS*. 2012;131(1):e109-e115.

16. Sharma R. Craniosynostosis. *Indian J Plast Surg*. 2013;46(1):18.

17. Turk AE, McCarthy JG, Thome CH, Wisoff JH. The "Back to Sleep Campaign" and deformational plagiocephaly. *J Craniofac Surg*. 1996;7(1):12-8.

18. Safe to Sleep [Internet]. Rockville: National Institute of Child Health and Human Development [cited 2021 October 10]. Progress in reducing SIDS. Available from: https://safetosleep.nichd.nih.gov/activities/SIDS/progress.

19. Fontana SC, Daniels D, Greaves T, Nazir N, Searl J, Andrews BT. Assessment of deformational plagiocephaly severity and neonatal developmental delay. *J Craniofac Surg*. 2016;27(8):1934—6.

20. Fowler EA, Becker DB, Pilgram TK, Noetzel M, Epstein J, Kane AA. Neurologic findings in infants with deformational plagiocephaly. *J Child Neurol*. 2008;23(7):742—7.

21. Collett BR. Development in toddlers with and without deformational plagiocephaly. *Arch Pediatr Adolesc Med*. 2011;165(7):653.

22. Speltz ML, Collett BR, Stott-Miller M, Starr JR, Heike C, Wolfram-Aduan AM, et al. Case-control study of neurodevelopment in deformational plagiocephaly. *PEDIATRICS*. 2010;125(3).

23. Miller RI, Clarren SK. Long-term developmental outcomes in patients with deformational plagiocephaly. *PEDIATRICS*. 2000;105(2).

24. Roby BB, Finkelstein M, Tibesar RJ, Sidman, JD. Prevalence of Positional Plagiocephaly in Teens Born after the "Back to Sleep" Campaign. *Otolaryngol Head Neck Surg*. 2012;146(5):823—8.

25. Robinson S, Proctor M. Diagnosis and management of deformational plagiocephaly. *J Neurosurg Pediatr.* 2009;3(4):284—95.

26. Boere-Boonekamp MM, van der Linden-Kuiper LT. Positional Preference: Prevalence in Infants and Follow-Up After Two Years. *PEDIATRICS*. 2001;107(2):339—43.

27. Hutchison BL, Hutchison LA, Thompson JM, Mitchell EA. Plagiocephaly and brachycephaly in the first two years of life: a prospective cohort study. *PEDIATRICS*. 2004;114(4):970-80.

28. Hutchison BL, Stewart AW, Mitchell EA. Deformational plagiocephaly: a follow-up of head shape, parental concern and neurodevelopment at ages 3 and 4 years. *Arch Dis Child*. 2011;96(1):85-90.

29. Collett B, Breiger D, King D, Cunningham M, Speltz M. Neurodevelopmental Implications of "Deformational" Plagiocephaly. *J Dev Behav Pediatr*. 2005;26(5):379—89.

30. Langlois J, Ritter J, Casey R, Sawin D. Infant Attractiveness Predicts Maternal Behaviors and Attitudes. *Dev Psych*. 1995; 31(3), 464-4.

31. Kapp-Simon K, Mcguire D. Observed Social Interaction Patterns in Adolescents with and without Craniofacial Conditions. *Cleft Palate Craniofac J*. 1997;34(5):380—4.

32. Shapiro D, Waljee J, Ranganathan K, Buchman S, Warschausky S. Using the Patient Reported Outcomes Measurement Information System to Evaluate Psychosocial Functioning among Children with Craniofacial Anomalies. *Plast Reconst Surg*. 2015;135(6):1673-9.

33. Bous R, Hazen R, Baus I, Palomo J, Kumar A, Valiathan M. Psychosocial Adjustments Among Adolescents With Craniofacial Conditions and the Influence of Social Factors: A Multi-Informant Study. *Cleft Palate Craniofac J*. 2019.

34. Margaret M, Walster E. The Effect of Physical Attractiveness on Teacher Expectations. *Sociol Educ*. 1973;46(2).

35. Jones A, Plumb AM, Sandage MJ. Social Media as a Platform for Cyberbullying of Individuals With Craniofacial Anomalies: A Preliminary Survey. *Lang Speech Hear Serv Sch*. 2021;52(3):840-8.

36. Cummings C. Positional plagiocephaly. *Paediatr Child Health*. 2011;16(8):493-6.

37. Mawji A, Vollman A, Robinson A, Tak F, Hatfield J, McNeil D, et al. Risk factors for positional plagiocephaly and appropriate time frames for prevention messaging. *J Paediatr Child Health*. 2014;19(8):423—7.

38. Ballardini E, Sisti M, Basaglia N, Benedetto M, Baldan A, Borgna-Pignatti C, et al. Prevalence and characteristics of positional plagiocephaly in healthy full-term infants at 8—12 weeks of life. *Eur J Pediatr*. 2018.

39. Vlieger A, Blink M, Tromp E, Benninga M. Use of complementary and alternative medicine by pediatric patients with functional and organic gastrointestinal diseases: results from a multicenter survey. *PEDIATRICS*. 2008;122(2):e446-e451.

40. Saedt E, Driehuis F, Hoogeboom TJ, van der Woude BH, de Bie RA, Nijhuis-van der Sanden MWG. Common manual therapy practices in the Netherlands for infants with upper cervical dysfunction: A prospective cohort study. *J Manipulative Physiol Ther*. 2018;41(1):52—61.

41. Rubinstein S, Pfeifle CE, van Tulder MW, Assendelft WJJ. Chiropractic patients in the Netherlands: A descriptive study. *J Manipulative Physiol Ther*. 2000;23(8):557—63.

42. Fludder C, Keil B. Deformational plagiocephaly and reduced cervical range of motion: a pediatric case series in a chiropractic clinic. *Altern Ther Health Med*. 2020;15.

43. Fincham JE. Response rates and responsiveness for surveys, standards, and the journal. *Am J Pharm Educ*. 2008;72(2):43.

44. Lee ACC, Li DH, Kemper KJ. Chiropractic care for children. *Arch Pediatr Adolesc Med*. 2000;154(4):401.

45. Durant CL, Verhoef MJ, Conway PJ, Sauve RS. Chiropractic treatment of patients younger than 18 years of age: Frequency, patterns and chiropractors' beliefs. *J Paediatr Child Health*. 2001;6(7):433—8.

46. Hestbaek L, Stochkendahl MJ. The evidence base for chiropractic treatment of musculoskeletal conditions in children and adolescents: the emperor's new suit?. *Chiropr Osteopat*. 2010;18(1).

47. Verhoef M, Papadopoulos C. Survey of Canadian chiropractors' involvement in the treatment of patients under the age of 18. J Can Chiropr Assoc. 1999;43(1):50-7.

48. Hestbaek L, Jørgensen A, Hartvigsen J. A description of children and adolescents in Danish chiropractic practice: results from a nationwide survey. *J Manipulative Physiol Ther*. 2009;32(8):607—15.

49. Miller J. Demographic survey of pediatric patients presenting to a chiropractic teaching clinic. *Chiropr Osteopat*. 2010;18(1).

50. De Inocencio J. Musculoskeletal pain in primary pediatric care: analysis of 1000 consecutive general pediatric clinic visits. *PEDIATRICS*. 1998;102(6).

51. Davies N. Chiropractic management of deformational plagiocephaly in infants: an alternative to device-dependent therapy. *Chiropr J Aust*. 2002;32(2):52-5.

Infantile colic as the clinical expression of brainstem dysregulation: a narrative review

Jan Hoeve, PhD, DC
Chiropractie Staphorst, The Netherlands
janhoeve@chiropractiestaphorst.nl

ABSTRACT

Despite many years of research, the cause of infantile colic is as elusive as ever and no distinct cure has emerged. Evidence is reviewed that colic may be the clinical expression of physiological dysregulation at the brainstem level, particularly of the vestibular and autonomic systems. It is argued that occipital/upper-cervical muscular tightness acquired at birth may induce aberrant proprioceptive flow into the vestibular system and, indirectly, the medial cerebellar cortex. Diminished inhibitory modulation of the vestibular nuclei by the medial cerebellum may provide a mechanism of how vestibular dysregulation/ hyperactivity may arise and subsequently be propagated downstream to also involve the autonomic systems. Treatment aimed at relaxation of tight sub-occipital musculature may restore regular proprioceptive flow and facilitate normalization of the inhibitory cerebellar modulation of the vestibular nuclei. From an evolutionary perspective the high prevalence of infantile colic may be an inherent aspect of the human condition and may be the price we pay for our upright stance, bipedal gait and difficult birth process as compared to the great apes. The focus of future research should be broadened to include the concomitant symptoms of colic that point to brainstem involvement.

Key Words: Infantile colic, brainstem dysregulation, sub-occipital dysfunction, proprioception, sensory neuromodulation, vestibular, autonomic.

Introduction

With an estimated prevalence of 15-25% infantile colic is one of the commonest early childhood afflictions, along with Functional Gastro-Intestinal Disorders (FGID)[1,2] Still, colic remains enigmatic. To date, 68 years after the rule-of-three diagnostic criteria were formulated[3] and after a great deal of research, the cause of colic is as elusive as ever and no distinct cure has emerged. In the intervening years much research has been focused on the crying per se and on establishing if and how colicky crying differs from normal crying[2,4] In addition, a multitude of possible contributing factors, pathological and non-pathological, have been proposed, often in a rather haphazard fashion. These include dietary causes such as cow's milk or fructose intolerance, GER(D), (neuro)developmental problems, behavioral problems, difficult temperament, transient hyperresponsivity, immaturity of the gut, gut dysbiosis, over/ underfeeding, imbalance of the autonomic nervous system, high levels of distress.[5,6,7] Organic pathology is rarely present.[8] Several authors have suggested that colic is just the extreme end of a spectrum of normal crying behavior,[9,10] although this is unlikely from an evolutionary as well as an energy perspective.[11] Others pointed to the dynamics of infant-parent interaction.[12] This is also unlikely as it cannot account for those cases where one baby of a pair of twins is colicky whereas the other is not.[11] In recent years a close association with childhood/adolescent migraine has become apparent whose significance, however, is still uncertain.[13]

After all these years, the diagnosis is still one of exclusion, being based upon excess crying in the absence of a recognizable cause. This puzzling lack of progress may suggest one of two possibilities, either colic is such an intractably difficult problem that 68 years was simply too short an interval for a solution to be found, or the research effort may have been misdirected.

From a biological perspective, small infants are primitive creatures who still function at a very basic level of visceral brainstem reflexes, because higher inhibitory modulating structures are not sufficiently developed yet.[14,15] This implies that any behavioral or physiological dysregulation observed may reasonably be expected to arise from this same level. Yet, the possibility that infantile colic could reflect a dysregulation at the brainstem level has received scant attention.

In a recent study conducted in my clinic, a 5-point clinical index of vestibular (hyper)activity was presented and applied as a tool to evaluate brainstem dysregulation in colicky babies before and after gentle treatment compared to non-colicky babies (see information box next page).[11] Colicky babies, it appears, are not just babies who cry a lot. They also show clinical evidence of vestibular dysregulation i.e., dysregulation at the brainstem level. Before treatment, the vestibular index was 7.8 times higher for colicky than non-colicky babies. Following treatment, the index had decreased by 96.5%, signifying a normalization of vestibular regulation.

In this review, relationships between infantile colic and

brainstem dysregulation are further investigated, as is the role of upper-cervical muscular dysfunction acquired at birth.

Colic and brainstem dysregulation

As one of the first sensory systems to emerge in the embryological development of vertebrates, the vestibular nuclei have extensive projections to other brainstem nuclei and play a central role in many regulatory processes.[16,17] This means that any vestibular dysregulation present may be propagated downstream to also involve these other brainstem nuclei, particularly those of the autonomic and the trigeminal systems.[18,19] Indeed, from scattered sources throughout the literature, supportive evidence of brainstem dysregulation in infantile colic can be gleaned. Though little studied, concomitant symptoms that are commonly associated with colic include (1) asymmetric posture (C-curve) and head preference even while asleep, which may lead to developmental plagio/brachiocephaly;[20] (2) extensor hypertonicity and pseudo-opisthotonic posture;[20,21] (3) upper-cervical movement/joint dysfunction, muscular tightness and occipital tenderness;[20] (4) high levels of stress and stress arousal;[9,21,22] (5) (breast)feeding difficulties;[23-26] (6) gastro-intestinal regulatory disorders such as regurgitation /Ger(d) and intestinal cramps.[1]

Extensor posture

Relaxed, happy babies have a snug, flexion-dominated fetal posture.[14] Their necks are wobbly and when held upright against the parent's chest the head needs to be supported. In stark contrast colicky babies are extension-dominated and arch their back.[20] Parents must be alert not to let the baby slip from their hands (back dive). These babies are noted to have a "strong neck," a "strong back" and "strong legs" and, when held in front, they try to climb up against the parent's chest. When placed in a prone position, they tend to lift their head, pull up their legs and try to move forward in a froglike fashion, even as early as the first few days after birth. Because in young infants, extensor posture and hypertonicity point to activation of anti-gravity muscles by the vestibulo-spinal tracts, the observed postural characteristics (1,2,3) are suggestive of vestibulo-spinal hyperactivity.[15,16,27]

The clinical index is based upon the consideration that mild rhythmic stimulation has a relaxing, soothing effect, whereas overstimulation tends to be uncomfortable and may lead to dizziness, nausea or even vomiting. If a baby does not react well to mild vestibular stimulation this is taken as a possible sign of vestibular (hyper)activity. The clinical index is comprised of five statements each of which can be answered by a simple agree/disagree. Each "agree" earns one point and each "disagree" earns zero points. These statements are based upon literature reports, personal observations and reported parental experiences:

(A). Your baby does not calm down or fall asleep during a car ride.

(B). Your baby does not calm down or fall asleep when held against your chest or cradled in the crook of your arm, while walking around at a brisk pace.

(C). When your baby has fallen asleep against your chest you cannot lay the baby supine in the crib without the baby waking up and crying.

(D). When sleeping the baby may wake up with a scream, showing the symptoms of the Moro reflex.

(E). The baby is much more comfortable lying inclined in a car seat than supine in a crib.

Information box 1: 5-point clinical index of vestibular (hyper) activity (reference 11)

Stress characteristics

Colicky babies are restless, irritable, in obvious distress, have trouble falling asleep and have fragmented sleep patterns (15-20 minute cat naps), they scratch their face, are easily startled, react strongly to minor sensory stimuli (hyperreactivity, hyperarousal, hypervigilance) and are easily overstimulated.[21] In addition, they tend to have cold clammy hands and feet. Together these symptoms point to sympathetic activation, as do the shrill and high-pitched acoustic characteristics of colicky crying.[22,25,28-33] Studies of covariation of infant cry acoustics and autonomic state have shown that the pitch of infant cries and other aspects of vocal prosody provide a sensitive index of autonomic activity.[34] Less vagal output is associated with increased pitch, whereas increased vagal output is accompanied by a lowering of pitch. Shrill high frequency cries indicate distress reflecting decreased vagal output or increased sympathetic activation. In a recent development a mobile telephone app ("Chatterbaby") incorporating a cry-translation algorithm allowed discrimination of cries associated with fussiness, hunger, pain and colic with 70-90% accuracy.[35] It was found that the cries of colic were similar to those of pain, but of higher intensity and pitch, suggesting that colic may be painful and stressful. Another study reported that increased levels of salivary cortisol in colicky babies were related to fragmented sleep patterns and intensity of crying, suggesting activation of the HPA-stress system.[36] Actually, the sympatico-adrenomedullary (SAM) and the hypothalamic-pituitary-adrenocortical (HPA) stress systems may both be activated.[24]

Feeding problems

Co-occurrences between infantile colic and feeding problems are reported, but interrelationships have been little studied. In hospital-admitted colicky babies, feeding problems were twice as common as in non-colicky babies.[24] In a different study colicky babies had (i) more gastro-esophageal reflux (GER); (ii) were less responsive during feeding interaction with the mother; (iii) had more episodes of feeding discomfort and (iv) had greater difficulty

coordinating sucking, swallowing and breathing.[23] This led the authors to suggest that colic and feeding problems could both be related to some underlying disorder of behavioral dysregulation. An association between feeding problems and colic was also reported in a study that looked at suck-swallow-breathing-coordination (SSBC) in colicky and non-colicky babies.[26] The authors observed that the duration of feeding in colicky babies was twice the normal 15-20 minutes in non-colicky babies. In addition, colicky babies showed dysfunctional relationships for elements of SSBC, similar to but more subtle than those observed in infants with neurological difficulties. Though in colicky babies there is no evidence of neurological deficit, the feeding difficulties could still reflect a dysregulation of brainstem autonomic nuclei, because sucking, swallowing, breathing and vocalization are coordinated by the nucleus ambiguus of the vagus system.[28,37]

Gastro-intestinal problems

In small infants, gastro-intestinal disorders including regurgitation (gastro-esophageal reflux, GER) constipation and intestinal cramps with or without obstipation or dyschezia are common in association with colic.[1,38] Frequent associations are gas/bloating/colic, gas/bloating/regurgitation and regurgitation/colic. These symptoms have been interpreted as early manifestations of later childhood painful FGID's.[39-41] Regurgitation (Ger) involves a transient relaxation of the lower esophageal sphincter triggered by gastric stretch receptors and not by passive mechanical distension of the gastro-esophageal junction.[42,43] This may indicate that excessive regurgitation could reflect dysregulation of autonomic neural control. Alternatively, the infants could just be nauseous due to dysregulation and hyperactivity of the vestibular system.[11,19] In case of constipation the presence of dry hard stools may suggest decreased gut motility due to decreased parasympathetic output or sympathetic activation and point to autonomic dysregulation.[44] Infant dyschezia is defined as excessive straining for more than 10-20 minutes with or without passing of stools[1] and is thought to be due to failure to coordinate intra-abdominal pressure with relaxation of the muscles of the pelvic floor.[45] Again, this may point to dysregulation of autonomic neural control.

Colic, upper-cervical dysfunction and mild birth trauma

Scattered evidence suggests that the origin of the upper-cervical muscular dysfunction and occipital tenderness may be related to the birthing process. One study documented an association between colic and feeding problems and found that the main risk factor was a complicated pregnancy or birth process.[24] Another study reported that breast feeding problems were more common following assisted births than non-assisted births.[46] Stressed and difficult temperamental behavior apparently are already present during the first few days after birth,[47] and high responsivity (stressed) behavior during the first week was predictive of colicky behavior during the following weeks.[48] A recent MRI study revealed that already shortly after birth "colicky babies showed greater sensitivity to olfactory stimuli than their non-colicky peers."[49]

Biedermann[20] was the first to have specifically linked upper-cervical dysfunction and colic to mild birth trauma related to the difficult human birth process as compared to the great apes. In the course of human evolution, the development of bipedal locomotion and the concomitant adaptation of the pelvic architecture preceded the increase in brain size by several million years.[50] In modern humans there is a very tight fit between the size of the baby and the maternal birth canal leaving only a small margin of error. As a result, the human baby must negotiate a narrow and tortuous birth canal[51-53] leading to a high level of birth complications with mother and child in comparison to the great apes.[54] During birth the skull is subjected to molding forces while the neck undergoes a considerable degree of rotation and extension.[55,56] It is easy to envisage how in a percentage of births such twisting may result in upper-cervical movement and joint dysfunction, muscular tightness and tenderness,[20] particularly in view of the observation that small meningeal bleeds are common even in non-symptomatic neonates.[57,58] In most births the final exit turn is to the right, and, accordingly, if there is a distinct head preference, in 70-80% of cases this is also to the right.[59] In chimpanzees with their easier birth process inconsolable crying as in human colicky babies is unknown.[60] Although infant chimps cry, they stop crying as soon as they are handled by the mother.

Colic, brainstem dysregulation and upper-cervical dysfunction

How these entities are interlinked may be inferred from the realization that upper-cervical proprioception plays

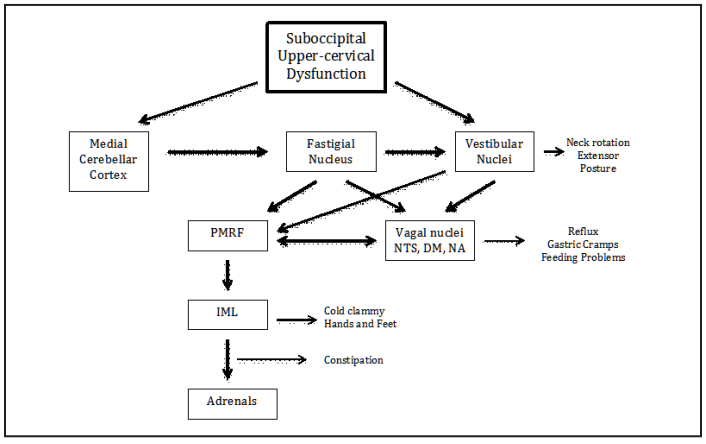


Figure 1: Functional relationships of sub-occipital musculature, medial cerebellar nuclei, vestibular nuclei, reticular formation, vagal nuclei and adrenals. PMRF — pontomedullary reticular formation; NTS — nucleus tractus solitarius; DM — dorsal motor nucleus; NA-nucleus ambiguus; IML — intermediolateral cell column.

an important role in modulating the vestibular system, as does the visual system.[61-63] Afferent proprioceptive input from upper-cervical segments (C1-3) and from axial structures project to the vestibular nuclei and indirectly to the medial cerebellar cortex and its purkinje cells and from there to the fastigial nucleus, which is the output nucleus of the medial cerebellum (Figure 1).[16] By inhibition of the fastigial nucleus the purkinje cells of the medial cerebellum indirectly exert an inhibitory modulating influence on the vestibular system.[16,19,27,64] Vestibular hyperactivity may arise even in case of weak, subclinical, diminished cerebellar inhibition.[19,65] Consequently, aberrant proprioceptive traffic into the vestibular system and the medial cerebellum, arising from tight sub-occipital musculature acquired at birth could conceivably lead to dysregulation and hyperactivity of the vestibular nuclei. Conversely, treatment achieving relaxation of the tight musculature may be expected to restore regular proprioceptive flow and facilitate normalization of the inhibitory cerebellar modulation of the vestibular nuclei.

Young infants in particular may be vulnerable to vestibular dysregulation, because compensation via the visual system would not be available as visual gaze and the vestibulo-ocular reflex (VOR) remain to be fully established.[15] In small babies, vestibular modulation may be almost entirely dependent on upper-cervical proprioception. The vestibular and the fastigial nuclei send efferent projections to the autonomic nuclei and to the reticular formation, and from the latter onwards to the sympathetic chain and the adrenal glands (Figure 1).[16,66,67] This means that dysregulation of the vestibular nuclei may propagate along these projections to also induce dysregulation of the sympathetic and parasympathetic systems, such as inferred in the above review of concomitant symptoms.

Colic and childhood/adolescent migraine

Although a close association between colic and migraine seems well established,[13,68-70] the discussion of its significance is hampered by the fact that there is no generally agreed pathophysiological model for either of these disorders. Some authors have suggested that colic and migraine may share a common pathogenic mechanism such as excessive sensitivity to sensory stimuli[71] or increased sensitization of perivascular nerve endings in the meninges and the gut,[13] possibly mediated by calcitonin-gene-related peptide (CGRP). At least one case-control study found an association between migraine and functional gastro-intestinal disorders in children and adolescents.[72] Circadian rhythms have also been suggested as being at the base of both colic and migraine.[71] Other authors have suggested that there may be a continuum from colic, as the earliest manifestation of migraine, through other childhood precursors of migraine, to migraine in adolescents and adults.[73]

Discussion

It is proposed that in a percentage of births the human baby may suffer mild trauma, as initially suggested by Biedermann,[20] and that this may have far reaching effects.[11] The associated muscular tightness and accompanying movement/joint dysfunction may have an adverse effect on the vestibular and autonomic systems of the brainstem. Via a mechanism of aberrant proprioception, this may lead to dysregulation/ hyperactivity of the vestibular nuclei and the ensuing concomitant autonomic symptoms of colic. Such a mechanism may explain why in many babies the full spectrum of colicky behavior is not reached until several weeks after birth.[9,10] The finding of a 96.5% improvement of the vestibular index following treatment consisting of mild sensory stimulation[11] is consistent with restoration of regular proprioceptive flow and normalization of vestibular modulation by the medial cerebellum. It would suggest that central neuromodulation may be accomplished not only by cutaneous vibratory stimulation of the cymba of the external ear (sensory neuromodulation),[74] but also by similar proprioceptive stimulation of the sub-occipital musculature, at least in babies.[11] The spontaneous resolution of colicky behavior after 3-5 months may be a consequence of developing cortical control and inhibitory modulation of brainstem reflexes during this same period and may, thus, represent a developmental aspect of the disorder.[15,37]

However, this is not to say that in untreated babies the underlying dysfunction also resolves in all cases as there is accumulating evidence of longtime sequelae.[75,76] In a 10-year prospective study an association was found between infantile colic and recurrent abdominal pain, allergic disorders, sleep disorders, fussiness, aggressiveness.[77] A second study found that at three years of age former colicky babies had more sleeping problems and more frequent temper tantrums than former non-colicky babies. Also, families with colicky babies had more distress three years later.[76] In a third study ex-colicky babies at four years of age were more emotional and had more temper tantrums and more complaints of stomachache.[78] Infantile colic has also been linked to an increased risk of Irritable-Bowel-Syndrome (IBS) after four years of age.[41] By contrast, treated colicky babies three years later were less likely to experience long-term sequelae than colicky babies who had not been treated.[79]

As to the nature of the link between infantile colic and migraine, there is a tendency to not only consider colic as the earliest age-specific manifestation of migraine, but also implicitly equate colic with early migraine.[70,73] Some authors even went so far as to suggest that migraine medication could be useful in controlling colic.[68] The relationship, however, cannot be that straightforward, because the great majority of colicky babies do not develop migraine.

If colic is to be an early manifestation of migraine, this

would probably only apply to a minority of genetically predisposed individuals. In the only available cohort study, 23% of ex-colicky babies developed migraine, 77% did not and of the ex-non-colicky babies 11% still did.[80] From twin studies it is estimated that 30-60% of migraine cases are accounted for by genetics, which leaves a remaining 70-40% to non-genetic external and environmental factors.[81] If, as argued in this review, babies' function at the basic level of visceral reflexes and if infantile colic is the clinical expression of brainstem dysregulation, it would stand to reason that the link with childhood migraine should also be sought at this level. Speculatively, and from an evolutionary perspective, the high 15-25% prevalence of infantile colic may be an inherent aspect of the human condition and could be part of the price we pay for our upright stance, bipedal gait, and the difficult way we are born.

Future research

Given that after all those years the diagnosis of infantile colic is still one of exclusion, it is paramount that criteria be developed for a diagnosis based on distinct and objective clinical observations. A first step towards this end has been made by the earlier study conducted in my clinic in which a 5-point clinical index of vestibular hyperactivity was presented and applied to evaluate brainstem dysregulation in colicky babies before and after treatment versus non-colicky babies.[11] This index offers the prospect for development into a tool for a positive diagnosis. The recognition of the various concomitant symptoms reviewed above allows additional clinical indices to be developed i.e., postural index, stress index, index of feeding behavior, regurgitation index, index of gastric cramps. Once confirmed and validated, these indices together with the vestibular index may be integrated into a comprehensive instrument toward an objective and practical clinical diagnosis of infantile colic. It is suggested that the focus of future research should be broadened to also include the associated symptoms that are indicative of brainstem dysregulation. Also, the link with childhood/adolescent migraine should be further explored.

Conclusion

Colicky babies are much more than just infants who cry a lot. They also show clinical evidence of dysregulation at the brainstem level, particularly of the vestibular and autonomic nuclei. As a hypothesis it is proposed that colicky behavior, vestibular dysregulation and concomitant autonomic dysregulation are linked to sub-occipital/ upper-cervical muscular dysfunction secondary to mild birth trauma. Treatment aimed at relaxing tight sub-occipital/ upper-cervical musculature (sensory neuromodulation) may correct aberrant proprioceptive outflow to the vestibular nuclei and the medial cerebellum. This may facilitate normalization of vestibular inhibitory modulation by the cerebellum and lead to improvement of brainstem regulation. It would suggest that central neuromodulation can be effectuated, not only by vibratory sensory stimulation of the auricular branch of the vagus nerve, but also by similar proprioceptive stimulation of sub-occipital/upper-cervical muscular structures, at least in babies.

Authors contribution

The author is solely responsible for all aspects of this publication.

Funding

There was no external funding.

Competing interests

The author is director and owner of the clinic Chiropractie Staphorst.

References:

1. Benninga, M.A., et al., Childhood Functional Gastrointestinal Disorders: Neonate/Toddler. *Gastroenterology*. 2016 150(6): p. 1443-1455.e2.

2. Wolke, D., Bilgin, A. and Samara, M., Systematic Review and Meta-Analysis: Fussing and Crying Durations and Prevalence of Colic in Infants. *J Pediatr*. 2017;185: p. 55-61 e4.

3. Wessel, M.A., Cobb, J.C., Jackson, E.B., Harris, G.S. and Detwiler, A.C., 1954; Paroxysmal fussing in infancy, sometimes called colic. *Pediatrics*. 2017;14(5): p. 421-35.

4. Vermillet,A.Q, Tolboll, K., Litsis Mizan, S., Skewes, J.C. and Parsons, C.E., Crying in the first 12 months of life: A systematic review and meta-analysis of cross-country parent-reported data and modeling of the "cry curve". *Child Dev* 2022 93(4): 1201-1222. doi: 10/1111/cdev.13760.

5. Savino, F., Focus on infantile colic. *Acta Paediatr*, 2007; 96(9): p. 1259-64.

6. Johnson, J.D., Cocker, K. and Chang, E., Infantile colic: Recognition and Treatment. *Am Fam Physician*, 2015, Oct1; 92(7): 577-82.

7. Sung, V. 2018; Infantile colic, *Aust Presc*. 2007;41(4): 105-110. doi:10.18773/austprescr.2018.033.

8. Freedman, S.B., Al-Harthy, N. and Thull-Freedman, J. The crying infant: diagnostic testing and frequency of serious underlying disease. *Pediatrics*. 2009;123(3): p. 841-8.

9. St James-Roberts, I., 1989; Persistent crying in infancy. *J Child Psychol Psychiatry*, 2009; 30(2): p. 189-95.

10. Barr, R.G., The normal crying curve: what do we really know? *Dev Med Child Neurol*, 1990; 32(4): p. 356-62.

11. Hoeve, J. Clinical evidence of vestibular dysregulation in colicky babies before and after chiropractic treatment vs. non-colicky babies. Front. Pediatr. 2021; 9: 668457. doi:10.3389/fped.2021.668457.

12. Raiha, H., Lehtonen, L., Huhtala, V., Saleva, K. and Korvenranta, H., Excessively crying infant in the family: mother-infant, father-infant and mother-father interaction. *Child Care Health Dev*, 2002; 28(5): p. 419-29.

13. Gelfand, A.A., K.C. Thomas, and P.J. Goadsby, Before the headache: infant colic as an early life expression of migraine. *Neurology*, 2012. 79(13): p. 1392-6.

14. Jouen, F., 1992; Head position and posture in newborn infants. In: *Head-neck sensory motor system*, 2002; Berhoz, A., Graf W. and Vidal P. (Eds); Oxford University Press.

15. Sarnat, H.B., Functions of the corticospinal and corticobulbar tracts in the human newborn, *Journal of pediatric neurology*. 2003; 1(1):3-8,

16. Beck, R., 2008; *Functional neurology for practitioners of Manual Therapy*: 2003; Churchill Livingstone. ISBN-13: 978-043-10220-2,

17. Sailesh, G.K.S., Ravikanth, M. and Kumar, A.H.S., *J Med Sci Health* 2018; 4(2):1-10. doi:10.46347/jmsh. 2018.v04i02.001.

18. Wilkinson, D., et al., Preventing Episodic Migraine With Caloric Vestibular Stimulation: A Randomized Controlled Trial. *Headache*, 2017; 57(7): p. 1065-1087.

19. Bernetti, L., et al., Subclinical vestibular dysfunction in migraineurs without vertigo: A Clinical study. *Acta Neurol Scand*, 2018; 138(4): p. 270-277.

20. Biedermann, H., Manual therapy in children: proposals for an etiologic model. *J Manipulative Physiol Ther*. 2005; 28(3): p. e1-15.

21. Levine, M.I., 1956; Emotional factors in the etiology of colic in infants. *Pediatrics*. 2005; 18(5): p. 836-8

22. Fuller, B.F., Keefe, M.R. and Curtin, M. Acoustic analysis of cries from "normal" and "irritable" infants. *West J Nurs Res*, 1994; 16(3): p. 243-51; discussion 251-3.

23. Miller-Loncar, C., Bigsby, R., Wallach, M. and Lester, B., Infant colic and feeding difficulties. *Arch Dis Child*, 2004; 89(10): p. 908-12.

24. Zwart, P., Vellema-Goud, M.G. and Brand, P.L. Characteristics of infants admitted to hospital for persistent colic, and comparison with healthy infants. *Acta Paediatr*, 2007; 96(3): p. 401-5.

25. Douglas, P.S. and Hill, P.S. 2013; A neurobiological model for cry-fuss problems in the first three to four months of life. *Med Hypotheses*, 2007; 81(5): p. 816-22.

26. Degenaar, H. and Kritzinger, A. 2015; Suck, swallow and breathing coordination in infants with infantile colic. *S Afr J Commun Disord*, 2007; 62(1): p. e1-e10.

27. Pompeiano, O., 1972; Vestibulospinal relations: vestibular influences on gamma motoneurons and primary afferents. *Prog Brain Res*, 2007; 37:p 197-232. doi: 10.1016/S0079-6123(08)63904-7.

28. Porges, S; Cardiac vagal tone: a physiological index of stress, *Neurosci Biobehav Rev.*; 1995; 19(2):225-33; doi:10.1016/0149-7634(94)00066-a.

29. Lester, B., Boukydis, C.Z., Garcia-Coll, C., Hole, W. and Peucker, M., 1990; Infantile colic: Acoustic cry characteristcs, maternal perception of cry, and temperament 1992. *Infant behavior and Development*, 1995; 15, 15-26. doi: 10.1016/0163-6383(92)90003-O.

30. Zeskind, P.S. and Barr, R.G., Acoustic characteristics of naturally occuring cries of infants with "colic" *Child Development*, 1997; 68(3), 394-403.

31. Giddens C.L., Barron, K.W., Byrd-Craven, J., Clark, K.F. and Winter, A.S., Vocal indices of stress: a review; *J Voice*. 2013; 27(3):390.e21-9. doi:10.1016/j.jvoice.2012.12010. Epub 2013 Feb 23.

32. Jovancevic, S., Bolfan-Stosic, N. and Jovancevi, M., Differences in the acoustic characteristics of the cries of infants with colic and the cries of healthy infants; *Croatian Journal of Education*, 2014; 16(Sp.Ed.No2/2014):67-77.

33. Sondhi, S., Khan, M., Vijay, R., Salhan, A.K. and Chouhan, S., Acoustic analysis of speech under stress; *Int J Bioinform Res Appl*. 2015; 11(5):417-32. doi: 10.1504/ijbra.2015.071942.

34. Stewart et al, Covariation of acoustic features of infant cries and autonomic state. *Physiol Behav*. 2013 Aug 15; 120: doi 10.1016/j.physbeh.2013.07.003.

35. Parga et al, 2020; Defining and distinguishing infant behavioral states using acoustic cry analysis: is colic painful? *Pediatric research* 2013; 87, 576-580. doi.org/10.1038/s41390-019-0592-4.

36. Brand, S., Furlano, R.I., Sidler, M.A., Schulz, J. and Holsboer-Trachsler, E., "Oh, Baby, Please don't Cry": In infants suffering from Infantile Colic Hypothalamic-Pituitary-Adrenocortical Axis Activity is Related to Poor Sleep and Increased Crying Intensity. *Neuropsychobiology* 2011;64:15-23. doi:101159/000322456.

37. Porges and Fuhrman, The early development of the autonomic nervous system provides a neural platform for social behavior: A polyvagal

perspective. *Infant Child Dev*. 2011 Feb; 20(1):106-118. doi: 10.1002/icd.688.

38. Bellaiche, M., Oozeer, R., Gerardi-Temporel, G., Faure, C. and Vandenplas, Y., Multiple functional gastrointestinal disorders are frequent in formula-fed infants and decrease their quality of life. *Acta Paediatr*. 2018 Jul; 107(7): 1276-1282. doi: 10.1111/apa.14348. Epub 2018 apr 26.

39. Partty, A., Kalliomaki, M. Salminen, S. and Isolauri, E., Infant Distress and Development of Functional Gastrointestinal Disorders of Childhood: Is there a connection? *Jama Pediatr*. 2013; 167(10): 977-978. doi:10.1001/jamapediatrics.2013.99.

40. Indrio F., Di Mauro, A., Riezzo, G., Cavallo, L. and Francavilla, R. Infantile colic, regurgitation, and constipation: an early traumatic insult in the development of functional gastrointestinal disorders in children? *Eur J Pediatr*. 2015 Jun; 174(6): 842-2. doi:10.1007/s00431-014-2467-3. Epub 2014 Dec 13.

41. Kim et al, Infantile Colic and the Subsequent Development of the Irritable Bowel Syndrome, *J Neurogastroenterol Motil* 2022; 28(4): 618-629 doi.org/10.5056/jnm21181.

42. Penagini, R., Carmagnola, S., Cantu, P., Allocca, M. and Bianchi, P.A. Mechanoreceptors of the proximal stomach: Role in triggering transient lower esophageal sphincter relaxation. *Gastroenterology*, 2004 Jan;126(1):49-56. doi:10.1053/j.gastro.2003.10.045.

43. Massey, B.T., Simuncak, C., leCapitaine-Dana, N.J. and Pudur, S., Transient lower Esophageal Sphincter relaxations do not result from passive opening of the cadia by gastric distension. *Clinical-Alimentary tract*, 130, issue 1, p89-95, jan o1, 2006. doi: 10.1053/j.gastro.2005.11.003.

44. Liu, L., Milkova, N., Nirmalathasan, S. et al. Diagnosis of colonic dysmotility associated with autonomic dysfunction in patients with chronic refractory constipation. *Sci Rep* 12, 12051 (2022) doi:10.1038/s41598-022-15945-6.

45. Hyman, P.E., et al, 2006; Childhood Functional gastrointestinal disorders: neonate/toddler. *Gastroenterology*. 2006 Apr; 130(5): 1519-26. doi: 10.1053/j.gastro.2005.11.065.

46. Miller, P-12 Is there an association between birth type and area of musculoskeletalcomplaint in the neonate? In: ECU convention 2018 congress proceedings. *Chiropr Man Therap* 26, 44 doi.org/10.1186/s12998-018-0213-z.

47. Tsuchiya, H. Emergence of temperament in the neonate: neonates who cry longer during their first bath still cry longer at their next bathings. *Infant Behav Dev*. 2011 Dec; 34(4): 627-31. doi:10.1016/j.infbeh.2011.06.001. Epub 2011 Jul 12.

48. St James-Roberts, I., Goodwin, J., Peter, B. Adams, D. and Hunt, S., Individual Differences in responsivity to a neurobehavioural examination predict crying patterns of 1-week-old infants at home. *Developmental medicine & Child Neurology* 2003, 45: 400-407. doi:10.1017/S0012162203000756.

49. Adam-Darque, A et al, Shedding light on excessive crying in babies. *Pediatric Research*. 2020; 89(5):1239-1244, doi:10.1038/541390-020-1048-6.

50. DeSilva, J.M. and Rosenberg, K.R. 2017: Anatomy, Development, and Function of the Human Pelvis. *The Anatomical Record* 2020; 300:628-632. doi:10.1002/ar.23561.

51. Rosenberg, K. and Trevathan, W. 2002; Birth, obstetrics and human evolution. *BJOG*, 2020; 109(11): p. 1199-206.

52. Wittman, A. and Wall, L. The Evolutionary Origins of Obstructed Labor: Bipedalism, Encephalization, and the human Obstetric Dilemma. *Obstetrical and Gynecological Survey*, 2007; 62, Number 11,p 739 -748. doi:10.1097/01ogx.0000286584.04310.5c.

53. Buck, S., "The Evolutionary History of the Modern Birth Mechanism: Looking at Skeletal and Cultural Adaptations," Totem: *The University of Western Ontario Journal of Anthropology*: 2011; Vol. 19: Iss. 1, Article 7.

54. Pavlice, M., Romero, R. and Mitteroecker, P., Evolution of the human pelvis and obstructed labor: new explanations of an old obstetrical dilemma. *Am J Obst Gynecol*. 2020 Jan; 222(1):3-16. doi:10.1016/j.ajog.2019.06-043. Epub 2019 June 25.

55. Bamberg, C., et al., Human birth observed in real-time open magnetic resonance imaging. *Am J Obstet Gynecol*, 2012; 206(6): p. 505 e1-6.

56. Trevathan, W., Primate pelvic anatomy and implications for birth. *Philos Trans R Soc Lond B Biol Sci*, 2015; 370(1663): p. 20140065.

57. Looney et al, Intracranial hemorrhage in Asymptomatic Neonates: Prevalence on MRI Images and Relationships to Obstetric Risk Factors 1. February 2007. *Radiology* 2007; 242(2):535-41. doi:10.1148/radiol.2422060133.

58. Rooks et al, Prevalence and Evolution of Intracranial Hemorrhage in Asymtomatic Term Infants. *Am J of Neuroradiology* June 2008, 29(6) 1082-1089; doi:103174/ajnr.A1004.

59. Michel, G.F. and Goodwin, R., Intrauterine Birth Position predicts Newborn supine Head position Preference. *Infant Behavior and Development*, 1979: 2:29-38. doi:10.1016/ S0163-6383(79)80005-3.

60. Bard, K.A. Crying in infant primates: Insights into the development of crying in Chimpansees. In: Eds. Barr RG, Hopkins B and Green JA, Crying as a Sign, a Symptom & a Signal. *Clinics in Developmental Medicine* 2000; No 152, Chapter 10, p 157-175.

61. Gdowski, G.T. and McCrea, R.A. Neck proprioceptive inputs to primate vestibular nucleus neurons. *Exp Brain Res*, 2000; 135(4): p. 511-26.

62. Deshpande, N. and Patla, A.E. 2005; Postural responses and spatial orientation to neck proprioceptive and vestibular inputs during locomotion in young and older adults. *Exp Brain Res*, 2000; 167(3): p. 468-74.

63. Sprenger, A., Wojak, J.F., Jandi, N.M. and Helmchen, C., Postural Control in Bilateral Vestibular Failure: Its Relation to Visual, Proprioceptive,

Vestibular, and Cognitive Input. *Front Neurol*. 2017; 8: p. 444.

64. De Zeeuw, C.I. and Berrebi, A.S. Postsynaptic targets of Purkinje cell terminals in the cerebellar and vestibular nuclei of the rat. *Eur J Neurosci*. 1995; 7(11): p. 2322-33.

65. Gerwig, M., Rauschen, L., Gaul, C., Katsarava, Z. and Timman, D., Subclinical cerebellar dysfunction in patients with migraine: Evidence from eyeblink conditioning. *Cephalgia* 24, 2014. doi:10.1177/0333102414523844.

66. Batton, R.R., Jayaraman, A., Ruggiero, D. and Carpenter, M.B., Fastigial efferent projections in the monkey: an autoradiographic study. *J Comp Neurol*, 1977; 174(2): p. 281-305.

67. Andrezik, J.A., Dormer, K.J., Foreman, R.D. and Person, R.J., 1984; Fastigial nucleus projections to the brain stem in beagles: pathways for autonomic regulation. *Neuroscience*, 1977; 11(2): p. 497-507.

68. Romanello, S. et al., Association between childhood migraine and history of infantile colic. *JAMA*, 2013. 309(15): p. 1607-12.

69. Sillanpää, M. and Saarinen, M., Infantile colic associated with childhood migraine: A prospective cohort study. *Cephalalgia*, 2015. 35(14): p. 1246-51.

70. Gelfand, A.A., Episodic Syndromes That May Be Associated With Migraine: A.K.A. "the Childhood Periodic Syndromes". *Headache*, 2015. 55(10): p. 1358-64.

71. Gelfand, A.A., Infant colic-a baby's migraine? *Cephalalgia*, 2015. 35 (14): p. 1243-5.

72. Le Gal, J. et al., Association between functional gastrointestinal disorders and migraine in children and adolescents: a case-control study. *Lancet Gastroenterol Hepatol*, 2016. 1(2):p. 114-121.

73. Epstein, L.G. and Zee, P.C., Infantile colic and migraine. *JAMA*, 2013. 309(15): p. 1636-7.

74. Addorisio, M.E., et al., 2019; Investigational treatment of rheumatoid arthritis with a vibrotactile device applied to the external ear. *Bioelectron Med*. 1977; 5: p. 4.

75. Wolke, D., Rizzo, P. and Woods, S., Persistent infant and hyperactivity problems in middle childhood. *Pediatrics*. 2002 Jun; 109(6):1054-60. doi: 10.1542/peds.109.6.1054.

76. Savino et al., 2005; A prospective 10-year study on children who had severe infantile colic. *Acta Paediatr Suppl*. 2005 Oct; 94(44):129-32. doi: 10.11/j1651-2227.2005, tb02169.x.

77. Rautava, P., Lehtonen, L., Helenius, H. and Sillanpaa, M., 1995; Infantile colic: child and family three years later. *Pediatrics*, 96(1 Pt 1): p. 43-7.

78. Canivet, C., Jakobsson, I. and Hagander, B. 2000; Infantile colic. Follow-up at four years of age: still more "emotional". *Acta Paediatr*, 1977; 89(1): p. 13-7.

79. Miller, J.E. and Philips, H.L. Long-term effects of infant colic: A survey comparison of chiropractic treatment and non-treatment groups. *J Manipulative Physiol Ther* 2009; 32: 635-638, doi: 10.1016/j.jmpt.2009.08.017.

80. Sillanpää, M. and Saarinen, M. Infantile colic associated with childhood migraine: A prospective cohort study. *Cephalalgia*, 2015. 35(14): p. 1246-51.

81. Mulder EJ, Van Baal C, GAist D, Kallela M, Kaprio J, Svensson DA et al. Genetic and environmental influences on migraine: a twin study across six countries. *Twin Res* 6(5): 422-431 (2003).

Manual therapy and probiotic supplementation for infant colic: an evidence-based clinical evaluation

Susanne Williams-Frey, DC, MSc[1] and Joyce E Miller, DC, PhD

[1]Corresponding Author Susanne Williams-Frey
Private practice Ebikon, Switzerland; Email: chiropraktik.ebikon@yahoo.com

ABSTRACT

Aim: To determine whether research evidence supports probiotic supplementation together with spinal manipulative therapy as a viable combination to significantly decrease infant colic. **Method:** Review and report findings of scientific papers that support the effectiveness of treatments with probiotics and spinal manipulative therapy for infant colic. **Result:** Considerable research suggests that probiotic supplementation could improve metabolites such as short chain fatty acids that have been shown helpful in regulation of a stress response. Both probiotics and manual therapy appear to improve functionality of the vagus nerve and parasympathetic nervous system to balance the autonomic nervous system, which could result in reduction in crying time for infants. Each treatment, individually has shown some benefits for infant colic in randomized controlled trials. Both remedies are considered safe. **Conclusion:** Research suggests there may be sufficient background to justify randomized trials utilizing a combination of probiotic supplementation and manual therapy for the treatment of infant colic. Since both treatments are considered safe and benefits have been shown, it may be appropriate for individual clinicians, faced with a colicky infant, to suggest to the parents a short clinical trial combining these two therapies.

Key Words: infant colic, probiotics, manual therapy, autonomic nervous system.

Introduction: Infant colic: Why is finding an answer important?

While infant colic (aka excessive crying of infancy) occurs in about 20% of all infants,[1] inconsolable and persistent crying is often insufficiently treated. One reason for this might be that the etiology of infant colic remains unknown as scientific proof of cause has not been recognized so far. Additionally challenging is that the infant cannot be asked what and where it hurts, nor are established criteria available. Hence, the diagnosis is determined by the clinician through exclusion of other disorders.

According to the Mayo Clinic Website:[2]

"The baby's care provider does a complete physical exam to identify any possible other causes for the baby's distress. The exam includes: Measuring the baby's height, weight and head circumference. Listening to the heart, lungs and abdominal sounds. Examining the limbs, fingers, toes, eyes, ears and genitals. Assessing reaction to touch or movement. And looking for signs of rash, inflammation, or other signs of infection or allergies." If all this examination results in no findings and the infant is thriving, the diagnosis of infant colic can be made.

Although the etiology of infant colic is unclear, its' impact on the families is not. These consequences of infant colic, particularly on the child, have been slow to attract research. One study found by looking at the duration of infant colic, that although it has been widely characterized as limited to the first 12 weeks of life, there is significant evidence that this is not the case, for at least 50-60% of children.[3,4] Not only did these children cry for more than three months, they also showed emotional problems up to eight years of age.[4] Each developmental phase places new demands on the infant's capacity to self-regulate.

This capacity depends on the structural and functional maturity of the brain and nervous system as well as the accumulated experiences, which have already been integrated.[5] Social capabilities like persistent eye contact, social smiling, and melodious cooing starts at ages two and three months of age.[6] This preverbal communication provides a framework for practicing reciprocal regulation of attention, positive affective arousal, and self-efficacy.[5]

In a Norwegian study it was found that infants, who had infant colic were significantly less developed at five years of age and internalized problems more at three and five years old than the previously non-colicky children.[7] They concluded that infant colic should be taken into account as a risk factor for development and behavioral problems within the first five years of a child's life.[7]

How does excessive crying of the newborn impact parents? Infant colic affects between 17 and 25% of newborns during the first months of life, with a reported peak of excessive crying somewhere between three and six weeks of age.[8,9] The provisional number of births in the United States in 2020 was 3,605,201,[10] which would mean that between 612,884

and 901,300 baby's families were dealing with a colic infant in the US alone. These families were most likely in distress, often not only because the excessively crying child could not be consoled, but also had other issues. For example, in many cases, the colicky infant is having problems in sleep-wake organization as well.[5,7] And this may keep parents awake during sleep hours. Hence, they then must cope with increasing sleep deprivation, while trying to function in everyday life tasks. This can cause parents to be overwhelmed and in psychosocial distress. Furthermore, this could be followed by frequent interactional failure with the child which consequently maintains or exacerbates behavioral problems,[11] and therefore increases infant crying.

Frequent emotional effects of inconsolable crying on parents causes the exhibition of signs of chronic exhaustion and overload, due to persistent alarm and sleep deficit. Repeated daily experience of infant inconsolability induces feelings of failure, diminished self-esteem, powerlessness, and depression.[5] Almost all affected parents confess that feelings of helplessness in the face of increasing arousal and alarm occasionally turn into a state of aggressive feelings and powerless rage toward the infant. These impulses may in turn evoke intense feelings of guilt and make the parents increasingly vulnerable. The intensity of aroused feelings may finally become fertile ground for the revival of latent conflicts with the partner or other family members, or over the mother's abandoned professional career.[5]

Moreover, excessive crying is significantly associated with shaking and smothering the child.[12] Shaken baby syndrome or abusive head trauma is the leading cause of death due to child abuse.[13] The mortality rate from shaken baby syndrome has been reported as 20-25%.[14,15] Further, even in the non-fatal cases, the shaken baby syndrome has a very poor outcome and major long-standing negative consequences are frequent.[16] In its minimal form, shaken baby syndrome consists of subdural hematoma. In 75 to 90% of cases, the subdural hematoma is associated with uni- or bilateral retinal hemorrhage.[14,17,18] Therefore, if the shaking does not lead to death, it often leads to permanent handicap.[19]

Remedies or methods for treating infant colic are therefore urgently needed because it puts newborn babies and their care takers at risk for a tremendous amount of distress. Additionally, infant colic may inhibit the newborn from feeling comfortable in this world and developing a feeling of deep trust. Family bonds are disrupted when the infant is most vulnerable and needs it desperately.[20] Furthermore, the child is at risk for parental abuse, which could lead to permanent damage or even death.[14,17-19] Disadvantages do not stop there; other outcomes might be developmental and behavioral problems within the first five years of a child's life.[4,7]

"Stress diathesis" models suggest that adversity in early life alters the development of neural and endocrine systems in a manner that an individual becomes predisposed to disease in adulthood.[21] In an animal study, mother-infant interactions in the first few days of life were categorized into high and low intensity contacts. When the pups entered adulthood the researchers looked at developmental, behavioral and endocrine responses to stress and found that the pups that had more motherly contact when newly born were significantly less fearful under conditions of novelty than the ones that had fewer interactions.[22] That study (though done on animals, not humans) suggests that the quality of interactions by the caregiver toward the offspring can "program" behavioral and neuroendocrine responses to stress in adulthood or vulnerability/resistance to stress-induced illness over the lifespan.[22]

In the case of a colicky infant, interactions between the caregiver and the child are disrupted. A child cannot focus on or enjoy a caregiver's attention when distressed to the point of excessive crying. Playfull and cuddly interactions can only happen in a state of calm. In a study where feeding problems in infants with colic were assessed, it was found that the colicky infants did not only have more feeding problems but also were less responsive towards their mothers during feeding interactions than infants in the non-colic group. Regarding parent functioning, mothers in the colic group reported higher levels of stress as compared to mothers in the comparison group.[23] An overwhelmed as well as psychosocially distressed caregiver most likely cannot give as much care and affection as they would like, despite their love for their child. A healthy connection is disrupted and might even be perceived by the child as rejection. Ultimately this results in more stress as well as negative psychological as well as physiological responses years later.[7,22]

What treatments show promise and why?

In a systematic review[24] it was shown that the strongest evidence for the treatment of infant colic were probiotics, particularly Lactobacillus reuteri (L. reuteri). Unfortunately, this was the case for breastfed infants only.[25] However, in another scientific investigation it was found that Bifidobacterium breve (B. breve) showed efficacy in both breastfed and formula fed infants.[26] The second strongest evidence for an effective treatment of infant colic was manual therapy.[24] Probiotic supplementation and manual therapy both carry a very low risk of serious adverse events but have not shown consistent effectiveness in a sufficient number of trials to be widely adopted.[24]

These two most effective treatment options lead to the question: "Would efficacy increase if the two treatments were combined?" To answer this question, it is key to search for evidence for the underlying mechanisms of each

treatment. What could be the link between these different treatment approaches, and can they support or even augment each other and therefore lead to a scientifically effective treatment? It has been suggested that one cause for the infant to cry in excess might be a dysfunctional nervous system in particular the autonomic nervous system (ANS) with the vagus nerve being one of the main actors.[27] This would mean that in order to improve infant colic, regulation of the function of the ANS would be the primary therapeutic focus. Other researchers have suggested that an unbalanced gastrointestinal microbiome, increased intestinal permeability, and chronic inflammation are involved.[1] Compiling all these findings and investigating how these factors interconnect might answer the question as to why probiotic supplementation and manual therapy could be more effective together.

ANS interaction in the excessively crying infant: What is the connection?

The ANS, consisting of the sympathetic and parasympathetic nervous systems, controls and regulates functions of various organs like the gut, glands, and involuntary muscles throughout the body (e.g., vocalization, swallowing, heart rate, respiration, gastric secretion and intestinal motility).[27,28] A respected source states that one of the most important roles of the parasympathetic nervous system is to oppose the activity of the sympathetic nervous system in order to keep balance between them.[27] The parasympathetic connection between inner organs like the gut and the brain (gut-brain-axis) is formed by the vagus nerve as the main contributor. This leads to the vagus nerve being an important functional as well as rapid connection between the CNS and the enteric nervous system (ENS) of the gut.[27,28] The ENS consists of a nerve plexus embedded in the intestinal wall, which extends across the whole gastrointestinal tract from the esophagus to the anus. The estimated number of neurons is between 100-500 million and constitutes the largest single nerve cell collection in the human body. It is also called "the second brain" because of its similarities in structure, function, and chemical determinants with the brain.[27] The ENS afferent fibers in the abdominal vagus trunk outnumber efferent fibers by about 10 to 1.[29] Therefore, the vagus nerve can be considered more a sensory than a motor nerve, which conveys a vast amount of sensory information to the brainstem.[29]

The framework of the ENS is laid during the first gestational trimester, but the network continues to undergo modifications throughout the prenatal period and into postnatal life[30] and its cells arise from the same cells as the vagus nerve.[27] Colonization of the gastrointestinal tract by trillions of microorganisms during the early postnatal period represents a significant change from the prenatal condition that undoubtedly affects the developing ENS and consequently the vagus nerve. It has been suggested that the early microbiome supports the development of the ENS and therefore probiotics could have further potential in clinical implications.[30]

It is the anatomy of the vagus nerve that ties different etiologies and symptomatology together and might explain why supporting gut health and vertebral joint function could lead to a viable treatment combination. The vagus nerve originates in the brain, in the medulla oblongata of the brainstem. Parasympathetic efferent nerve fibers primarily go to the gastrointestinal tract, heart and lungs, but also to the muscles of the soft palate, pharynx and larynx. Primary afferent fibers come from visceral organs, including taste from the tongue as well as pain, temperature and deep touch of the outer ear, the dura of the posterior cranial fossa and the mucosa of the larynx.[31] From the brain the vagus nerve exits the skull laterally through the jugular foramens together with the accessory nerve (CN XI). The vagus nerve then passes distally in very close proximity to the transverse processes of the Atlas (C1) and Axis (C2) between the carotid artery and the internal jugular vein, within the carotid sheath — directly behind the sternocleidomastoid muscle and just anterior to the scalenes.[32] At the base of the neck, the nerve enters the thorax and the right and left vagus nerve diverge after this point. The left travels into the esophagus and the right to the right bronchus.[31] Both left and right vagus nerves subsequently enter the abdomen through the diaphragm and branches are sent to the esophagus, the stomach and primarily to the intestinal tract — up to the splenic flexure of the large colon, forming the ENS.[31]

A malfunctioning ANS or hyperactive sympathetic nervous system may be especially challenging to the newborn. This is because the human infant is not born with a completely functioning and myelinated vagal system. Therefore, its development continues in the first few months postpartum through activation. For the external observer, maturation of the vagal system can be distinguished by the increased speed of how efficiently an infant calms after a disruptive challenge and the increasing time period the infant remains calm.[33] For example, a non-colicky infant can be soothed by being carried or touched, but this has no impact on the excessively crying infant and sometimes even worsens their symptoms, as though these children have a unique threshold or delayed response to downregulation. Interestingly, an increase in sympathetic activity has consistently been found in several studies (in adults) with crying. After resolution of crying, the parasympathetic nervous system was activated.[34] Crying however is dependent on the functional integrity of the cranial nerve X (vagus).[35] Notably, the colicky infant does not calm down even when crying for hours or after feeding, nor does the colicky infant remain calm on a sufficiently increasing level. All these findings might show that the difference in the maturation of the

somatosensory processing pathways might be leading to the different responses or abilities to calm down between the colicky infant and non-colicky infant.[36] This leads to the question, what if the colicky infant is trying to activate the vagus nerve or parasympathetic nervous system through crying, but cannot?

Additionally, a lack of neuronal activation to and in the brain might affect brain development (factors detected in late effects of infant colic). Disturbances in the fine-tuning of interactions between myelination and functional connectivity maturation could disrupt some developmental processes. By compromising this neural network, it could be said that in the case of infant colic, an over excited sympathetic or insufficiently stimulated parasympathetic nervous system or an underdeveloped vagus nerve may be one of the underlying causes or contributing factors. Also, it can be suggested that the link between the gastrointestinal dysfunctions and brain induced stress behaviors in infant colic is the gut-brain axis formed mainly by the vagus nerve. Furthermore, an immature vagal system might impair brain development and therefore cause developmental delay. In turn, it could be hypothesized that strengthening the vagus nerve function, as the main parasympathetic actor and neural connection of the gut-brain-pathway,[27,28] might lessen symptoms of excessive crying and its sequelae.

How might probiotics affect infant colic? What probiotic might be most beneficial?

Study findings of the monospecies probiotic Lactobacillus reuteri (L. reuteri) have shown in four double-blind trials,[25] involving 345 infants with colic, that the active treatment group had less crying and/or fussing time than the placebo group. However, significant intervention effects have only been shown in exclusively breastfed infants but were insignificant in formula fed infants. In the treatment group the mean crying and/or fussing duration was reduced by 21 minutes by day seven and 25.4 minutes by day 21.[25] In these trials, infant colic was either defined by the Wessel's (crying > 3 hours per day, for > 3 days per week, for > 3 weeks) or by the modified Wessel's (crying > 3 hours per day, for > 3 days per week) criteria.[38] This would mean that the average breast-fed colic baby was still crying and/or fussing a minimum of two hours and 35 minutes at day 21 of receiving L. reuteri. Even if it is common for infants to cry about one hour per day by 10-12 weeks old,[8] two and one-half hours a day in a comparable time frame, is more than double the normal crying time. Therefore, although L. reuteri showed some effect in breastfed babies, it could not be considered a scientifically validated "cure."

Further, in a later study it was discovered that the combination of Lactobacillus rhamnosus (L. rhamnosus) and L. reuteri was more effective in alleviation of colic in breastfed infants than individually. The statistically significant difference between the probiotic and control groups was -47 minutes.[39] The monospecies L. rhamnosus showed no effect in the nonhomogeneous group of breast- and formula fed infants.[40] Because Infants who received combined L. reuteri with L. rhamnosus, were all exclusively breastfed, the role of L. rhamnosus is difficult to ascertain, although there could be some type of synergistic effect since the reduced crying time was increased with a combination of both species.[39]

In a study where B. breve was used, effectiveness was also shown in formula fed infants.[26] So, would a multispecies probiotic with the addition of B. breve be a more effective choice to treat infant colic in breastfed and formula fed infants? Breast milk contains multiple and beneficial health enhancing microbes such as Bifidobacterium subspecies (B. spp). Multiple studies have reported that breastfed infants have a higher abundance of beneficial B. spp. compared with formula fed infants.[41,42] Furthermore, in a recent review, where infants who received either breast or formula milk were compared, they found that feeding type modulates microbiome composition.[43] Moreover, they found that breastfed infants' fecal samples not only consisted of higher amounts of B. spp. but also of Lactobacillus spp. (L. spp.) and contained fewer pathogens.[43] In another study, in which the microbiomes of infants who received extensively hydrolyzed or amino acid formula were compared with infants receiving human milk, it was found again, that the amount of B. spp. was higher in breastfed infants.[44] The predominant presence of B. spp. in breast milk could have accounted for the fact that supplementation of L. reuteri has shown improvement with breastfed infants only.[25] When the two (L. spp. and B. spp.) are working in synchronicity, infant colic could improve in the formula fed colic infants too.

Just mixing B. and L. spp. together will not lead to an effective probiotic treatment. A probiotic formula needs to be carefully chosen, containing powerful human microbial strains from the area of treatment with scientific relevance. Therefore, a considered probiotic supplement needs to focus on individual properties of the bacterial strains, identity, safety as well as technological issues, such as stability and targeted release. Against this background, an example study report was written for a gynecological application, where 127 presumptive lactobacilli isolates of vaginal origin were collected. A step-by-step selection was done meeting specific criteria like compatibility and growth enhancement, which finally lead to a preparation consisting of four individual L. strains that possess particular significance in women's urogenital health.[45]

Evidence for why B. spp. most likely play an important role in early life is that they are the dominant bacteria in a healthy newborn gut microbiome.[46] One major function of the

Bifidobacterium genus is to contribute to gut homeostasis and host health with the involvement in the production of short-chain fatty acids (SCFA).[47,48] In fact, it was found that SCFA production is increased with increased amounts of B. spp.[47,49] Study findings showed that SCFAs, such as butyric acid, have a positive direct effect on vagal afferent terminals situated in the gut mucosa, which ultimately helps - through the gut-brain-axis — with the regulation of brain function.[27,50,51] Additionally, it was shown in a study that breastfed infants displayed lower inflammatory cytokine profiles than formula fed infants.[52] This might be partially due to breast milk containing significant amounts of B. spp. and therefore SCFAs such as butyrate. Butyrate can regulate gut permeability by reducing pro-inflammatory cytokines and increase anti-inflammatory cytokine circulation.[53] This in turn decreases inflammatory stress to the infant's body. On the other hand, it may be considered that formula fed infants do have a higher tendency for inflammatory processes to occur, and therefore could experience more dysfunctions in the gut and the brain as a consequence of diminished amounts of B. spp.

How are B. spp. introduced to the baby's gut? In a study where meconium of newborns was analyzed, it was found that the number of bacterial strains increased with time after birth and accumulated at an estimated rate of 1.2 strains per day.[54] The earliest species detected were facultative anaerobes (can live with and without oxygen) from the Enterobacteriaceae and Bacilli, notably Streptococcus and Enterobacter. First obligate anaerobes (die in an oxygen environment) were detected after 25 hours, including Clostridia, whereas B. spp. were found more than 100 hours after birth,[50] which indicates that breastfeeding, or external components, like oral (retrograde) translocation,[55] introduces the newborn to B. spp. Another external source to introduce the newborn to B. spp. could be supplementation. A study has shown that administration of certain probiotic strains of bacteria (mainly B. spp. and L. spp.) can positively alter the metabolic profile of the host through production of neurogenic metabolites that positively influence the nervous system including the brain, as well as support an anti-inflammatory environment.[27,47]

A dysbiosis or insufficient amount of SCFA or lactic acid producing bacteria, like B. spp. could lead to an increased number of inflammatory metabolites, which then could cause a malfunctioning neural system including the brain. Furthermore, evidence has shown that an increased risk for immune disorders like allergies and infections may result.[43,47,52] Hence, it could be argued that inflammation as well as dysbiosis might be decreased with a probiotic containing B. spp. In fact, B. spp. might be the deciding factor in a probiotic formula to reduce discomfort and crying in the formula fed colicky infant. Not only the formula fed infant could benefit, but also the breastfed colicky infant, because supplementation with a multispecies probiotic containing B. spp. could increase the speed of gut microbiome and ANS balance. However, formula fed or breastfed, based on study results, the colicky infant could benefit from a multispecies probiotic containing B. spp. by a reduction of aggravating factors. Moreover, studies have shown that commensal gut bacteria strengthen vagal function and therefore gut-brain-axis interaction.[27]

Probiotic supplementation may correct a dysbiotic microbiome, which in turn changes the metabolite composition and causes an increase in production of SCFAs, neurotransmitters, like serotonin as well as anti-inflammatory cytokines such as Interleukin[10,47,56] Further, this change in metabolite composition results in a decrease of inflammation and improvement in gut health, as well as a stress response like crying.[48,49] Additionally, vagus nerve endings in the intestinal wall (ENS) are activated by bacterial metabolites. With proper bacterial metabolite production, the vagus nerve is stimulated and its function as well as its development improves. Consequently, parasympathetic input from the gut to the brain and in the brain is increased,[27] which may improve brain maturation and therefore decrease the potential for developmental delay.[36,37] All these findings could support a randomized trial using a multi-species probiotic with B. as the leading probiotic bacteria.

How might manual therapy affect infant colic?

Within the general population, manual therapy is an often sought alternative therapy by parents of an unsettled, fussy and irritable infant.[57] Practitioners working with colicky infants have experienced repeatedly, that inconsolable crying can disappear after only a few treatments. Unfortunately, scientific evidence has not been able to prove significant effectiveness through a sufficient number of randomized controlled trials. One reason is that randomized controlled trials are difficult to develop for newborns and they are quite costly as trials are in general, which makes funding a challenge. Further, studying this subject has been difficult because of varying definitions and measurements in an already low number of studies. Additionally, it is commonly believed (though not proven and increasingly unlikely) that infant colic is a self-limiting disorder. In a recent systematic scoping review, it was therefore concluded that findings for the effectiveness of spinal manipulation to manage infant colic for crying time and sleep disturbances were inconclusive.[58]

That said, there are randomized trials that show statistically significant improvement in cry times with SMT.[59] Further, anecdotal manual therapy successes are encouraging and may have some value. Why else would so many parents seek this treatment option?[57] Is it simply a parental placebo response? Based on a randomized controlled study done

by Miller et al.[59] where the crying time reduction was statistically significant at day eight and was not susceptible to parental bias, the answer would be no. In their study it was also observed that the drop out rate in the non-treatment group was significantly higher, suggesting that parents didn't find the trial helpful and began looking for other types of care.[59] Considering that it is the parents that spend the most time with the infant, this observation might be valuable.

Accordingly, in an article by Hughes and Bolton[60] it was determined: "The evidence suggests that chiropractic has no benefit over placebo in the treatment of infantile colic. However, there is good evidence that taking a colicky infant to a chiropractor will result in fewer reported hours of colic by the parents. And therefore, in a clinical scenario where the family is under significant strain, where the infant may be at risk of harm and possible long term repercussions, where there are limited alternative effective interventions, and where the mother has confidence in a chiropractor from other experiences, the advice is to seek chiropractic treatment."

It has not been adequately explained why manual therapy seems to decrease or alleviate symptoms of infant colic. It has been proposed that the ANS might play a role.[57] The complexity of the ANS becomes apparent when it fails to function. Because a complex system is constructed of multiple connections, it becomes a challenge when trying to track it back to its origin, which is a prerequisite for correction. On the other hand, a complex system is a multipathway system and compensatory routes may be necessary. Therefore, in the event of malfunction, critical systems like the musculoskeletal and nervous system will still be maintained, except in a state of compromised and maladaptive autonomic neuroplasticity.[61] Further complicating the impact of malfunctioning of the ANS is that it is conceptualized as the intermediary between the human internal and external environments (i.e.,vagus nerve afferent), whereas the brain is optimizing adaptation to internal and external stressors (i.e., vagus nerve efferent).[62] A faulty or dysregulated ANS might therefore not be able to access the external environment correctly and consequently the brain cannot sufficiently adapt to stressors.

Malfunctioning of the vagus nerve, which innervates structures in the head, neck, thorax and abdomen, could therefore change physiological responses of cells, tissues, organs and systems in these areas. Fortunately, adaptive systems are activated or inhibited efficiently, but not excessively, so that the human body can cope with these challenges. However, there are several processes in which body systems either are overstimulated or inadequate in their response to stimulation. It is the cost of adaptation that may trigger pathophysiology.[63] A recent study highlighted that modulation of the ANS has been a mechanism underlying the interventions in complementary and integrative techniques such as manual therapy,[61] which supports the notion that chiropractic manipulation could counteract a malfunctioning central nervous system (CNS) or ANS.[64]

In the case of the colicky infant, their systems are still in the developmental stage. At this early stage synchronization of operating systems is limited and drawing from sufficient resources to compensate may be lacking. Body systems in a newborn have just begun to incorporate gained experiences, which are very sparse at this point. This is especially critical when examing the capacity of the immature nervous system. Furthermore, integration of external experiences is limited because in the excessively crying child, body systems are occupied in coping with internal aggravations. Internal hyperactivity in the nervous system most likely causes a state of constant fright and flight, which creates an unpleasant tumult. This turmoil may not be counter acted, even when crying for hours, because the parasympathetic nervous system is not able to rise to the challenge. What occurs in the infant will sooner or later be transported to the empathic care giver, which may in turn create more aggravation and ends in the entire family being overwhelmed.

How might spinal manipulative therapy (SMT) work with infant colic? It has been proposed that when the position of the head migrates forward — as, for example, with prolonged prenatal constraint - increased strain is placed on the muscles and ligaments of the head, neck, and shoulders. This abnormal head posture could result in altered joint positions and ultimately dysfunction, which then leads to abnormal neuronal afferent information to the brain and body.[65] Considering the anatomical close proximity of the cranial nerves V to XII to the cervical spine and the brainstem, it would make sense, that an increase in adverse mechanical tension in the cervical spine may unfavorably act on the brainstem and cranial nerves V through XII. Hence, dysfunctions of the upper cervical spine could be one of the mechanisms that decreases parasympathetic input to or from the brain. Restoration of cervical function should then result in a balanced ANS. In a study by Moustafa et al. it was found, that improved cervical function improved sympathetic skin response as well as reduced longitudinal stress and strain on the cervical elements.[65] Another review showed that SMT, by stimulating the vagal nerve, improved autonomic imbalance.[64] It has been found that one of the main key factors leading to these improvements is a network of neurophysiological connections between the cervical spine mechanoreceptors and the ANS.[65] This suggests that not only in theory spinal corrections balance the ANS and reduce nerve interference, but also in practice, manual therapy may manifest a shift away from sympathetic dominance.[65]

Intuitively it is often assumed that the excessive crying of the infant stems from discomfort. At birth, newborns are equipped with basic connections involved in pain processing, but major maturation and organization of their pain control networks occurs postnatally. Maturational differences in pain network could be related to specific patterns of sensitivity and regulation observed in crying of colicky infants compared to non-colicky infants. Therefore, excessive crying in infants could be due to differential pain thresholds, pain perception, and duration of response after painful stimuli.[66] Since SMT has been shown to result in both local and regional pain reduction, as well as positively influence the CNS with a general reduction of pain sensitivity,[67] it becomes a viable choice of treatment for the newborn. This statement is supported by an experimental research project which showed that SMT influenced the incoming/ascending pain signals (local nociceptive input affecting dorsal horn excitability or temporal summation) and/or the excitability of the central pain regulating mechanisms.[67,68] Their research suggested that discomfort or an overexcited nervous system could be balanced with SMT.

Furthermore, in healthy individuals, acute stress triggers an increase in sympathetic activity, which often includes an increase in threshold, that is induced by descending inhibition. This indicates a bi-directional relationship, where the ANS not only reacts, but stress modulates ANS activity.[69] In this regulatory circuit, the brainstem plays a central role by connecting the cerebrum, the cerebellum and the spinal cord with each other. Through these connections the brainstem modulates the function of major systems like the cardiac, respiratory and gastrointestinal systems by sending vagal efferent information to these organs.[70] This is why vagus nerve stimulation has become a therapeutic avenue in several inflammatory or painful disorders such as musculoskeletal diseases.[71]

This could mean that in the case of infant colic a dysregulated CNS or ANS could be balanced by upregulating the vagus nerve function with SMT. In a 2020 study[64] a mechanism for how the activated vagus nerve (parasympathetic system) counterbalanced the activity of the sympathetic system was explained. Vagal stimulation releases neurotrophins including brain-derived neurotrophic factor and nerve growth factor. Brain-derived neurotrophic factor is an important neuronal growth factor that regulates neuronal maturation, neurogenesis, synaptic plasticity and survival.[64] Nerve growth factor acts as a modulator of the hypothalamic-pituitary-adrenal axis and therefore contributes to maintaining the neuroendocrine systems.[64] The neuroendocrine system controls the body's response to stress, meaning that if it functions correctly, a stress response can be terminated[36] in the infant when needs like hunger are well managed. Further research has found that infants who had chiropractic care for infant colic showed significantly fewer emotional and sleep problems as toddlers.[72]

How would these two treatments work together?

If in fact, excessive crying (aka infant colic) is at least partly due to an overactive sympathetic or underactive parasympathetic nervous system, where the vagus nerve is the main parasympathetic contributor,[27] then strengthening[28] or decreasing irritation to the vagus nerve could balance the ANS and therefore decrease a stress response like crying. Strengthening of the vagus nerve function can be achieved by metabolites which are produced by probiotic bacteria. Further, manual therapy in the cervical region, with mechanical strain on vagal structures is reduced, can result in proper function of the ANS. Combining these two therapy forms, which have already shown to be both safe and useful by themselves[24] by approaching the problem of an excessively crying infant from two different angles — at least in theory — could produce a reasonable working solution for testing. Both therapies have demonstrated some reductions in crying times. Is there any potential to trial them together to see if they could synergistically work together for more significant crying time reductions?

Summarizing and connecting research findings could answer how these two treatment methods might prove scientific efficacy when combined. Infants with colic are more responsive and can manifest increased reactivity, but they also have a diminished regulatory capacity. Infant colic seems to involve a regulatory capacity problem in addition to a reactivity disorder.[36] Probiotics increase neural function by activating afferent vagal fibers in the ENS, which send information to the brain. SMT on the other hand restores proper function of vagal activity at the cervical spine. By addressing the problem at different areas of the nervous system, the effects might be augmented for faster balance capabilities of the ANS and therefore faster calming down of the colicky infant from an excited state.

As for recommendation, what probiotic species combination should be used in a supplement for colic infants, it is difficult to say because only thorough scientific laboratory evaluation and testing as explained in the study done by Domig et al.[45] might give an evidence based combination. As no scientifically effective probiotic has statistically significant evidence as of today,[27] these authors suggest that L. spp.[25,39,40,43] and B. spp.[26,41-44] in combination might be most likely to improve the condition of infant colic in both breastfed and formula fed infants. Since no RCT has shown what species combination is the right one for all colic infants, families with a colic infant might be supported best by the clinician when using a multispecies probiotic with L. spp. as well as B. spp. in combination with SMT.

Conclusion

Both probiotics and manual therapy have shown some

benefits for the excessively crying infant. It should be considered that chiropractic care along with a multispecies probiotic combined may be more effective than each method by itself. If so, improvement should be seen in all infants, regardless of their feeding type. Not only should crying time decrease or become less intense, but also gut health and immune function should improve and most of all, the infant-parent relationship becomes a nourishing experience for both. The risk/benefit ratio suggests that further studies that combine these two types of therapies could be a reasonable way forward to help infants and their parents.

References:

1. Daelemans S, Peeters L, Hauser B, Vandenplas Y. Recent advances in understanding and managing infantile colic. *F1000Research*. 2018;7:1426. www.ncbi.nlm.nih.gov/pmc/articles/PMC6134333/pdf/f1000research-7-16265.pdf.

2. Mayo Clinic. Patient Care & Health Information/Disease & Conditions. Colic. 1998-2023 Mayo Foundation for Medical Education and Research MFMER. Colic - Diagnosis and treatment - Mayo Clinic.

3. Clifford TJ, Campbell MK, Speechley KN, Gorodzinsky F. Sequence of infant colic: evidence of transient infant distress and absence of lasting effects on maternal mental health. *Arch Pediatr Adolesc Med* 2002;156:1183-8.

4. Wolke D, Rizzo P, Woods S. Persistent infant crying and hyperactivity problems in middle childhood. *Pediatrics* 2002; 109:1054-60.

5. Kim JS. Excessive crying: behavioral and emotional regulation disorder in infancy. *Korean Journal of Pediatrics*. 2011;54(6):229-233.

6. Emde RN, Osofsky HJ. Bonding, humanism, and science. *Pediatrics*. 1983;72:749-750.

7. Valla L, Smastuent MC, Andenaes R, Misvaer N, Olbjorn C, Helseth S. Association between colic and sleep problems in infancy and subsequent development, emotional and behavioral problems: a longitudinal study. *BMC Pediatrics*. 2021;21-23.

8. Wolke D, Bilgin A, Samara M. Systematic Review and Meta/Analysis: Fussing and Crying Durations and Prevalence of Colic in Infants. *J Pediatrics*. 2017;185:55/61e4.

9. Vandenplas Y, Abkari A, Bellaiche M, Benninga M, Chouraqui JP, Cokura F, Harb T, Hegar B, Lifschitz C, Ludwig T, Miqdady M, de Morais MB, Osatakul S, Salvatore S, Shamir R, Staiano A, Szajewska H, Thapar N. Prevalence and health outcomes of functional gastrointestinal symptoms in infants from birth to 12 months of age. *J Paediatrics Nutr*. 2015;61(5):531-537.

10. Hamilton BE, Martin JA, Ostermann MJK. Births: Provisional Data for 2020. NVSS, Vital Statistics Rapid Release. 2021;12. https://www.cdc.gov/nchs/products/index.htm.

11. Wurmser H, Laubereau B, Hermann M, Papousek M, von Kries R. Excessive infant crying: often not confined to the first 3 months of age. *Early Human Development*. 2001;64:1-6.

12. Fujiwara T, Yamaoka Y, Morisaki N. Self-Reported Prevalence and Risk Factors for Shaking and Smothering Among Mothers of 4-Month-Old Infants in Japan. *Journal of Epidemiology*. 2016;26(1): 4-13.

13. Duhaime AC, Christian CW, Rorke LB, Yimmerman RA. Nonaccidental head injury in infants-the "shaken-baby syndrome." *N Engl J Med*. 1998;338(25):1822-9. doi:10.1056/NEJM199806183382507.

14. American Academy of Pediatrics Committee on Child Abuse and Neglect. Shaken baby syndrome: Rotational Cranial Injuries Technical Report. *Pediatrics*. 2001;108:206-210.

15. Barlow K, Thompson E, Johnson D, Minns R. The neurological outcome of non-accidental head injury. *Pediatr Rehabil*. 2004;7:195-203.

16. Laurent-Vannier A, Toure H, Vieux E, Brugel DG, Chevignard M. Long-term outcome of the shaken baby syndrome and medicolegal consequences: a case report. *Annals of Physical and Rehabilitation Medicine*. 2009;52:436-447. doi:10.1016/j.rehab.2009.03.001.

17. Johnson DL, Braun D, Friendly D. Accidental head trauma and retinal hemorrhage. *Neurosurgery*. 1993;33:231-234.

18. Kivlin JD, Simons KB, Lazority S, Ruttum MS. Shaken baby syndrome. *Ophthalmology*. 2000;107:1246-1254.

19. Jazavant S, Parr J. Outcome following subdural haemorrhages in infancy. *Arch Dis Child*. 2007;92:343-347.

20. Gabard-Durnam LJ, McLaughlin KA. Do Sensitive Periods Exist for Exposure to Adversity? *Biol Psychiatry*. 2019;85(10):789-791. doi:10.1016/j.biopsych.2019.03.975.

21. Broerman R. Diathesis-stress model. In: Zeigler-Hill V, Shackelford TK eds. Encyclopedia of Personality and Individual Differences. Springer International Publishing. 2018:1-3. doi:10.1007/978-3-319-28099-8_891-1.

22. Meaney MJ. Environmental programming of stress responses through DNA methylation: life at the interface between a dynamic environment and a fixed genome. *Dialogues Clin Neurosci*. 2005;7(2):103-123.

23. Miller-Loncar C, Bigsby R, High P, Wallach M, Lester B. Infant colic and feeding difficulties. *Arch Dis Child*. 2004;89:908-912. doi:10.1136/adc.2003.033233.

24. Ellwood J, Draper-Rodi J, Carnes D. Comparison of common interventions for the treatment of infantile colic: a systematic review of reviews and guidelines. *BMJ open*. 2020;10:e035405. doi:10.1136/bmjopen-2019-035405.

25. Sung V, D'Amico F, Cabana MD, Chau K, Koren G, Savino F, Szajewska H, Deshpande G, Dupont C, Indrio F, Mentula S, Partty A, Tancredi D. Lactobacillus reuteri to Treat Infant Colic: A Meta-Analysis. *Pediatrics*. 2018; 141(1):e20171811.

26. Pereira AR, Rodrigues J, Albergaria M. Effectiveness of probiotics for the treatment of infantile colic. *AJGP*. 2022;15(8). doi:10.31128/AJGP-07-21-6062.

27. Breit S, Kupferberg A, Rogler G, Hasler G. Vagus Nerve as Modulator of the Brain-Gut Axis in Psychiatric and Inflammatory Disorders. *Frontiers in Psychiatry*. 2018;9:44. doi:10.3389/fpsyt.2018.00044.

28. Howland RH. Vagus Nerve Stimulation. *Current Behavioral Neuroscience Reports*. 2014;1:64-73.

29. Hillsley K, Grundy D. Vagus Nerve. *Science direct: Encyclopedia of Gastroenterology*. 2004. http://www.sciencedirect.com/topics/neuroscience/vagus-nerve.

30. Mungovan K, Ratcliffe EM. Chapter 19 — Influence of the Microbiota on the Development and Functions of the "Second Brain"-The Enteric Nervous System. The Gut-Brain Axis. Dietary, Probiotic, and Prebiotic Interventions on the Microbiota. Academic Press. 2016:403-421.

31. Adams B. The Anterior Cranial Fossa. Website: TeachMeAnatomy. 2022. Anterior Cranial Fossa - Boundaries - Contents - TeachMeAnatomy: http://teachmeanatomy.info/head/areas/cranial-fossa/anterior/#:~:text=The%20anterior%20cranial%20fossa%20consists%20of%20three%20bones%3A,by%20the%20inner%20surface%20of%20the%20frontal%20bone.

32. Centeno C. How the Vagus Nerve Can Cause Neck Pain. Regenexx. 2020. http://regenexx.com/blog/vagus-nerve-and-neck-pain/.

33. Stephen W, Furman SA. The Early Development of the Autonomic Nervous System Provides a Neural Platform for Social Behavior: A Polyvagal Perspective. *Infant Child Dev*. 2011;20(1):106—118. doi:10.1002/icd.688.

34. Bylsma LM, Gracanin A, Vingerhoets AJJM. The neurobiology of human crying. *Clin Auton Res*. 2019;29(1):63-73.

35. Jürgens U. The neural control of vocalization in mammals: a review. *J Voice*. 2009;23:1—10.

36. Ratka A, Sutanto W, Bloemers M, de Kloet ER. On the role of brain mineralocorticoid (type I) and glucocorticoid (Type II) receptors in neuroendocrine regulation. *Neuroendocrinology*. 1989;50(2):117-23. doi:10.1159/000125210.

37 Wallois F, Routier L, Bourel-Ponchel E. Chapter 25 - Impact of prematurity on neurodevelopment. *Handbook of Clinical Neurology*. 2020;173:341-375. https://doi.org/10.1016/B978-0-444-64150-2.00026-5.

38. Wessel MA, Cobb JC, Jackson EB, Harris GS Jr, Detwiler AC. Paroxysmal fussing in infancy sometimes called colic. *Pediatrics*. 1954;14(5):421-433. [PubMed, Archives]

39. Gerasimov S, Gantzel J, Dementieva N, Schevchenko O, Tsitsura O, Guta N, Bobyk V, Kaprus V. Role fo Lactobacillus Rhamnosus (FloraActive™) 19070-2 and Lactobacillus reuteri (FloraActive™) 12246 in Infant Colic: A Randomized Dietary Study. *Nutrients*. 2018;10(12):1975. doi:10.3390/nu10121975.

40. Pärtty A, Lehtonen L, Kalliomäki M, Salminen S, Isolauri E. Probiotic Lactobacillus rhamnosus GG therapy and microbiological programming in infantile colic: a randomized, controlled trial. *Pediatric Research*. 2015;78(4):470-475.

41. Collado MC, Cemada M, Baüerl C, Vento M, Perez-Matinez G. Microbial ecology and host-microbiota interactions during early life stages. *Gut Microbes*. 2012;3:352-365. doi:10.416/gmic.21215.

42. Lee SA, Lim JY, Kim BS, Cho SJ, Kim NY, Kim OB, Kim Y. Comparison of the gut microbiota profile in breast-fed and formula-fed Korean infants using pyrosequencing. *Nutr Res Pract*. 2015;9:242-248. doi:10.4162/nrp.2015.9.3.242.

43. Dimitrakopoulou EI, Pouliakis A, Falaina V, Xanthos T, Zoumpoulakis P, Tsiaka T, Sokou R, Iliodromiti Z, Boutsikou T, Iacovidou N. The Metagenomic and Metabolomic Profile of the Infant Gut: Can They Be "Predicted" by the Feed Type?. *J children MDPI*. 2022;9:154. doi.org/10.3390/children9020154.

44. Kok CR, Brabec B, Chichlowski M, Harris CL, Moore N, Wampler JL, Vanderhoof J, Rose D, Hutkins R. Stool microbiome, pH and short/branched chain fatty acids in infants receiving extensively hydrolyzed formula, amino acid formula, or human milk through two months of age. *BMC Microbiology*. 2020;20:337. doi.org/10.1186/s12866-020-01991-5.

45. Domig KJ, Kiss H, Petricevic L, Viernstein H, Unger F, Kneifel W. Strategies for the evaluation of potential vaginal probiotics from human sources: an exemplary study. 2014;5(3):263-272. doi:10.3920/BM2013.0069.

46. Turroni F, Peano C, Pass DA, Foroni E, Severgnini M. Claesson MJ. Kerr C, Hourihane J, Murray D, Fuligni F, Gueimonde M, Margolles A, De Bellis G, Ventura M. Diversity of bifidobacteria within the infant gut microbiota. *PLoS ONE*. 2012. doi.org/10.1371/journal.pone.0036957.

47. Khokhlova EV, Smeianov VV, Efimov BA, Kafarskaia LI, Pavlova SI, Shkoporov AN. Anti-inflammatory properties of intestinal Bifidobacterium strains isolated from healthy infants. *Microbiol Immunol*. 2012;56:27—39. doi:10.1111/j.1348-0421.2011.00398.x.

48. Rivière A, Moens F, Selak M, Maes D, Weckx S, De Vuyst L. The ability of bifidobacteria to degrade arabinoxylan oligosaccharide constituents and derived oligosaccharides is strain dependent. *Appl. Environ Microbiol. Environ*. 2014;80:204—217. doi:10.1128/AEM.02853-13.

49. Scott KP, Antoine JM, Midtvedt T, van Hemert S. Manipulating the gut microbiota to maintain health and treat disease. Microb. Ecol. *Health Dis*. 2015;26:25877. doi: 10.3402/mehd.v26.25877.

50. Wang H, Lee I-S, Braun C, Enck P. Effect of Probiotics on Central Nervous System Functions in Animals and Humans: A Systematic Review. *J Neurogastroenterol Motil*. 2016;22(4):589—605.

51. Koopman FA, Chavan SS, Miljko S, Grazio S, Sokolovic S, Schuurman PR, Mehta AD, Levine YA, Faltys M, Zitnik R, Tracey KJ, Tak PP. Vagus nerve stimulation inhibits cytokine production and attenuates disease severity in rheumatoid arthritis. *Proc Natl Acad Sci USA*. 2016;113:8284-9. doi:10.1073/pnas.1605635113.

52. Goehring KC, Marriage BJ, Oliver JS, Wilder JA, Barrett EG, Buck RH. Similar to those who are breast fed, infants fed a formula containing 2'-fucosyllactose have lower inflammatory cytokines in a randomized controlled trial. *J Nutr*. 2016;146:2559-2566. doi:10.3945/jn.116.236919.

53. Paparo L, Nocerino R, Ciaglia E, Di Scala C, De caro C, Russo R, Trinchese G, Aitoro R, Amoroso A, Bruno C, Di Costanzo M, Passariello A, Mssina F, Agangi A, Napolitano M, Voto L, Della Gatta G, Pisapia L, Montella F, Mollica MP, Calignano A, Puca A, Canani RB. Butyrate as a bioactive human milk protective component against food allergy. *Allergy*. 2021;76:1398-1415. doi:10.1111/all.14625.

54. Bittinger K, Zhao C, Li Y, Ford E, Friedman ES, Ni J, Kulkarni CV, Cai J, Tian Y, Liu Q, Patterson AD, Sarkar D, Chan SHJ, Maranas C, Saha-Shah A, Lund P, Garcia BA, Mattei LM, Gerber JS, Elovitz MA, Kelly A, DeRusso P, Kim D, Hofstaedter CE, Goulian M, Li H, Bushman FD, Zemel B, Wu GD. Bacterial colonization reprograms the neonatal gut metabolome. *Nat Microbiol*. 2020;5(6):838-847. doi:10.1038/s41564-0200694-0.

55. Fernandez L, Langa S, Martin V, Maldonado A, Jimenez E, Martin R, Rodriguez JM. The human milk microbiota: origin and potential roles in health and disease. *Pharmacol Res*. 2013;69:1—10. doi:10.1016/j. phrs.2012.09.001.

56. Chung HJ, Sim JH, Min TS, Choi HK. Metabolomics and lipidomics approaches in the science of probiotics: a review. *J Med Food*. 2018;21:1086-1095. doi.org/10.1089/jmf.2017.4175.

57. Carnes D, Bright P, Brownhill K, Carroll K, Engel R, Grace S, Vogel S, Vaucher P. Crying Unsettled and disTressed Infants Effectiveness Study of osteopathic care (CUTIES trial): Pragmatic randomised superiority trial protocol. *International Journal of Osteopathic Medicine*. 2020;38:31-38. doi.org/10.1016/j.ijosm.2020.08.001.

58. Milne N, Longeri L, Patel A, Pool J, Olson K, Basson A, Gross AR. Spinal manipulation and mobilisation in the treatment of infants, children, and adolescents: a systematic scoping review. *BMC Pediatr*. 2022;22:721.

59. Miller J, Newell D, Bolton E. Efficacy of chiropractic manual therapy in infant colic: a pragmatic singel-blind randomised controlled trial. *J Manipulative. Physol Ther*. 2012; 35(8):600- doi:10.1016/j.jmpt.2012.09.010.

60. Hughes S, Bolton J. Is chiropractic an effective treatment in infantile colic? *Arch Dis Child*. 2002;86(5):382-4. doi:10.1136/adc.86.5.382.

61. Fornari M, Carnevali L, Sgoifo A: Single Osteopathic Manipulative Therapy Session Dampens Acute Autonomic and Neuroendocrine Responses to Mental Stress in Healthy Male Participants. *J Am Osteopath Assoc*. 2017;117(9):559-567.

62. Bakken AG, Axén I, Eklund A, O'Neill S. The effect of spinal manipulative therapy on heart rate variability and pain in patients with chronic neck pain: a randomized controlled trial. *Trials*. 2019;20(1):590. doi:10.1186/s13063-019-3678-8.

63. Kettner N, Sclocco R. Vagus nerve neuromodulation and chiropractic care. *Chiropractic Economics*. 2021; http://www.chiroeco.com/vagus-nerve-neuromodulation-and-chiropractic-care/.

64. Kiani AK, Maltese PE, Dautaj A, Paolacci S, Kurti D, Picotti PM, Bertelli M. Neurobiological basis of chiropractic manipulative treatment of the spine in the care of major depression. *Acta Biomed*. 2020;91(13): e2020006.

65. Moustafa I, Youssef ASA, Ahbouch A, Harrison D. Demonstration of Autonomic Nervous Function and Cervical Sensorimotor Control After Cervical Lordosis Rehabilitation: A Randomized Controlled Trial. *J Athl Train*. 2021;56(4): 427—436. doi:10.4085/1062-6050-0481.19.

66. Leuchter RH-V, Darque A, Hüppi PS. Brain Maturation, Early Sensory Processing, and Infant Colic. *JPGN*. 2013;57(1). doi:10.1097/01.mpg.0000441929.05684.78.

67. Coronado RA, Gay CW, Bialosky JE, Carnaby GD, Bishop MD, George SZ. Changes in pain sensitivity following spinal manipulation: a systematic review and meta-analysis. *J Electromyogr Kinesiol*. 2012;22(5):752—67.

68. Bialosky JE, Bishop MD, Robinson ME, Zeppieri G Jr, George SZ. Spinal manipulative therapy has an immediate effect on thermal pain sensitivity in people with low back pain: a randomized controlled trial. *Phys Ther*. 2009; 89(12):1292—303.

69. Hallman DM, Olsson EMG, von Schéele B, Melin L, Lyskov E. Effects of heart rate variability biofeedback in subjects with stress-related chronic neck pain: a pilot study. *Appl Psychophysiol Biofeedback*. 2011;36(2):71—80.

70. Valenza G, Sclocco R, Duggento A, Passamonti L, Napadow V, Barbieri R, Toschi N. The central autonomic network at rest: Uncovering functional MRI correlates of time-varying autonomic outflow. *Neuroimage*. 2019;15(197):383-390.

71. Courties A, Berenbaum F, Sellam J. Vagus nerve stimulation in musculoskeletal dieseases. *Joint Bone Spine*. 2021;88(3):105149. doi.org/10.1016/j.jbspin.2021.105149.

72. Miller J and Phillips H. Long term effects of infant colic: a survey comparison of chiropractic treatments and non-treatment groups. *Journal of Manipulative and Physiological Therapeutics* 2009; 32(8): 635-638.

73. Nocerino R, De Filippis F, Cecere G, et al. The therapeutic efficacy of Bifidobacterium animalis subsp. lactis BB-12® in infant colic: A randomised, double blind, placebo-controlled trial. *Aliment Pharmacol Ther*. 2020;51(1):110-120. doi:10.1111/apt.15561.

Treatment options for ankyloglossia: A research informed review

Benjamin James Gibbs[1] and Emily Norton, MSc, IBCLC[2]

[1]Mchiro, Year 4, AECC UC, Bournemouth, United Kingdom
[2]Newborn Feeding Clinic Lead Tutor. AECC UC, Bournemouth, United Kingdom
Corresponding author: Benjamin J Gibbs, email: bgibbs2019@outlook.com

ABSTRACT

Ankyloglossia is thought to impact important early infant milestones associated with development such as latching and breastfeeding as well as potentially affecting palate formation and airway patency. Not only is this issue important for infants and neonates, who are in a vulnerable and malleable state where function dictates structure, but its effect on breastfeeding can be devastating for both mother and infant. The clinical question then is, 'what are the best treatment options for infants with ankyloglossia?'. Although there are positive links between frenotomy procedures and outcomes, follow-up of frenotomy is vague, incomplete and inconclusive. Likewise, evidence is inconclusive for management options such as manual therapy to treat surrounding structures and areas of dysfunction in order to relax tethered oral tissues. Lack of conclusive evidence to determine a clear way forward in ankyloglossia treatment requires a deeper look and search for the best evidence available to assist parents and clinicians to make the best decision for their infant's treatment. The goal of this clinical research-based viewpoint is to shed light on the treatment of frenotomy to help determine the best treatment at the right time. This treatment plan may be unique to each infant.

Key Words: Ankyloglossia, tongue-tie, lingual frenulum, manual therapy, diagnosis tool, management pathway.

Background

The diagnosis of ankyloglossia in infants is not a new concept and has been considered throughout history as early as the 18th century.[1] The most common treatment is a frenotomy.[2] It is difficult to ascertain the number of infants born with ankyloglossia, as prevalence ranges in literature from 0.1% to 12%.[2-4] In the United Kingdom the number of infants undergoing frenotomy procedures was most recently collected in 2011. These figures suggested that 11.8 frenotomies were carried out per 1,000 children per annum.[5] It is possible that over-diagnosis may lead to unnecessary surgeries.

This clinically focused article aims to evaluate research and potential consequences of the frenotomy procedures on neonates, as well as to provide an opinion in the consideration of other types of treatments for ankyloglossia.

Introduction

The lingual frenulum (LF) is considered an embryological tissue which, during the fetal period attaches to the underside of the tongue and the floor of the mouth.[2] The initial function of this tissue is thought to maintain the lips and tongue in union with the cranial bones during fetal growth.[2] In 2019 Mills et al[6] published an infant cadaver study assessing the components of the LF. Their paper hypothesised that the function of the LF was likely to change throughout growth to an ultimate role of stabilization.[6] Mills et al[6] highlight key inconsistencies in anatomy with the current understanding of the topic claiming that the LF was composed of a fold of fascial tissue.[6] This is an important discovery when considering the present understanding and management regarding ankyloglossia (AG) in pediatric patients. Ankyloglossia is currently defined as a congenital anomaly characterised by a short LF.[7]

An abnormally short frenulum (concurrent with certain theories) has been linked to restrictions in tongue mobility.[8] Typically, AG has been associated with problems for both mother and infant. Studies investigating AG draw comparisons between a functionally restrictive frenulum and difficulties latching, breastfeeding (BF), teething, obstructive sleep apnea and even speech.[2,7,9]

There is no doubt of the importance of a mother's breast milk when feeding the infant for the first six months and up to two years of life and beyond.[10] Nutritional value gained from the mother not only provides the infant with necessary proteins, fats and sugars to support life, but also significantly contributes to the maturation of the gut.[11] It is considered a vital time in the maturation of the infant where the healthy bacterium of the microbiome is cultivated. This contributes to both the infant's ability to digest and to the emergence of their immune system, enabling the infant to combat disease effectively.[12,13]

Krol and Grossmann demonstrated the impact of BF on infant psychology.[14] This could impact further issues such as mother and infant bonding, which can also affect aspects of emotional regulation, with the emotional deficit in some

infants that fail to attach to the breast even described as crippling.[2] Recent studies, although unable to identify the exact mechanisms that link brain myelination and cognitive outcomes in BF individuals, highlight the importance of known nutrients including long-chain polyunsaturated fatty acids and other important myelin components to assist early neurodevelopment.[15]

In 2021, the Academy of Breastfeeding Medicine produced a position statement on AG in breastfeeding dyads.[16] This position statement considered that AG may significantly impede infant latching, suckling and breast milk transfer, while also impacting maternal nipple discomfort and trauma during BF.[16] This results in an increased risk of discontinuing BF.[17] An infant struggling to latch due to AG could suffer impaired development, especially at these key stages in early growth.[18,19] As a result of this, it is important to ensure that the infant has minimal obstacles regarding BF. This evidence supports the hypothesis that AG is a significant problem that merits treatment to enable normal function.

Prevalence

Hazelbaker[3] in 2017 suggested the prevalence of infants with AG can be up to 10%. However, other studies have reported varied prevalence ranging from 0.1% to 12%.[2-4] Males have a greater prevalence than females (0.62).[20] The figures for current corrective frenotomy procedures are illusive. In England the Department of Health produced an Infant and feeding profile from 2003-2011.[5] In this literature the number of corrective frenotomy procedures in infants less than one years of age was around 11.8 per 1000 with a total of 8,088 procedures altogether in the year 2010 to 2011 (see tables 1 and 2).[5] However, this is not strictly an accurate representation of the number of these procedures carried out today as this data only represented procedures carried out in Strategic Health Authorities. The figures, although the most recent, are also outdated, having been collected over ten years prior to the writing of this paper.

From 2007 to 2011 there was a significant rise of these procedures each year (see table 1).[5] After plotting a forecast for future values, it suggests that if numbers for procedures continue to increase at the same rate, by 2022, the predicted number of frenotomies carried out in England would have been around 30,000 (see table 3). This signifies nearly 50 in 1000 babies having the procedure completed in these healthcare institutions (see table 4). Current diagnosis errors are thought to affect prevalence as there are numerous diagnostic tests which have been poorly validated.[21]

Diagnosis

Signs, symptoms, observation and a functional assessment can lead to a diagnosis of a functionally restrictive frenulum. The presence of one or more of the manifestations in table 5 should lead to the exploration of the infant's mouth and assessment of the BF technique and nipples as well as a clinical breastfeeding assessment.[2]

Current diagnosis is determined with a combination of both identification of a 'short' lingual frenulum and diminished oral function; this is because the limit of function is not always impacted by the length of the LF. Geddes et al,[7] suggest this is because the function of infant sucking is affected by more components than just the tongue.

Assessment tools vary depending on location, institution and practitioner. The Hazelbaker assessment tool is

		2007/08			2008/09			2009/10			2010/11		
SHA	**Name**	**In Pat**	**Out Pat**	**Total**	**In Pat**	**Out Pat**	**Total**	**In Pat**	**Out Pat**	**Total**	**In Pat**	**Out Pat**	**Total**
Q30	North East HA	2.8	0.0	2.8	5.2	0.0	5.2	6.1	0.0	6.1	10.1	0.0	10.1
Q31	North West HA	1.1	0.6	1.7	1.8	0.7	2.5	2.5	1.6	4.1	2.6	9.2	11.7
Q32	Yorkshire and The Humber HA	0.9	0.8	1.7	1.5	1.3	2.8	2.0	2.0	4.0	2.6	3.8	6.4
Q33	East Midlands HA	0.6	0.2	0.8	0.7	0.9	1.6	0.9	1.7	2.6	1.5	3.7	5.2
Q34	West Midlands HA	1.1	1.6	2.7	3.0	3.0	6.0	4.9	5.4	10.4	5.9	6.2	12.1
Q35	East of England HA	3.9	0.0	3.9	4.7	0.2	4.9	6.9	2.2	9.0	6.8	2.5	9.3
Q36	London HA	2.1	0.0	2.2	2.8	0.1	2.9	3.5	5.2	8.7	6.0	5.4	11.3
Q37	South East Coast HA	1.7	1.4	3.1	3.4	2.9	6.3	6.5	12.0	18.5	6.1	14.3	20.3
Q38	South Central HA	2.1	0.3	2.4	2.7	1.4	4.2	2.7	1.4	4.1	5.1	2.7	7.8
Q39	South West HA	1.1	3.3	4.3	3.7	6.5	10.2	8.3	9.7	17.9	8.8	15.4	24.2
Q00	England	1.7	0.8	2.5	2.8	1.6	4.4	4.2	4.2	8.4	5.3	6.5	11.8

Table 1. Department of Health Infant Feeding Profile. Trend in number of inpatient & outpatient tongue tie (Ankyloglossia) corrective surgeries in infants aged less than 1 year old per 1,000 live births at Strategic Health Authorities in England.[5]

SHA	Name	2007/08			2008/09			2009/10			2010/11		
		In Pat	Out Pat	Total	In Pat	Out Pat	Total	In Pat	Out Pat	Total	In Pat	Out Pat	Total
Q30	North East HA	83	0	83	158	0	158	183	0	183	312	0	312
Q31	North West HA	95	51	146	157	66	223	222	138	360	230	817	1047
Q32	Yorkshire and The Humber HA	55	53	108	99	88	187	131	134	265	175	252	427
Q33	East Midlands HA	33	10	43	39	47	86	47	91	138	82	207	289
Q34	West Midlands HA	78	111	189	215	216	431	349	387	736	426	446	872
Q35	East of England HA	268	1	269	339	16	355	489	154	643	500	182	682
Q36	London HA	266	4	270	353	14	367	451	668	1119	794	716	1510
Q37	South East Coast HA	87	69	156	177	149	326	336	620	956	320	753	1073
Q38	South Central HA	109	14	123	144	76	220	139	73	212	275	145	420
Q39	South West HA	61	186	247	219	380	599	483	564	1047	532	924	1456
Q00	England	1,135	499	1,634	1,900	1,052	2,952	2,830	2,829	5,659	3,646	4,442	8,088

Table 2. Department of Health Infant Feeding Profile. Trend in number of inpatient & outpatient tongue tie (Ankyloglossia) corrective surgeries in infants aged less than 1 year old at Strategic Health Authorities in England.[5]

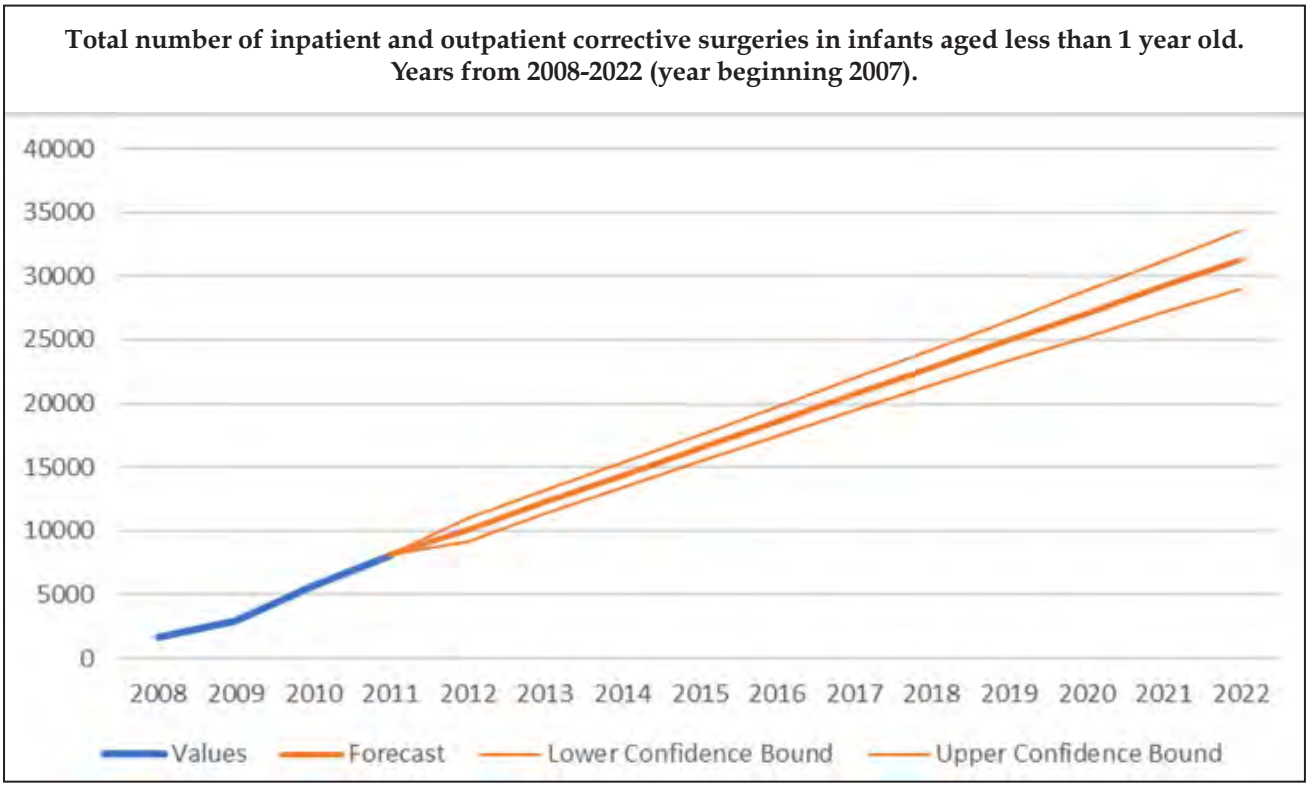


Table 3. Forecast for Trend in total number of inpatient and outpatient tongue tie (ankyloglossia) corrective surgeries in infants aged less than 1 year old at SHA's in England from the year beginning 2007.

commonly known.[22] This assesses five appearance and seven functional items of the tongue reviewing aesthetics and properties of the tongue, and how the tongue resides and is operational within the mouth of the infant.[23] The five appearance items consist of appearance when lifted, elasticity, length when tongue is lifted, attachment to tongue and attachment to inferior alveolar ridge (see table 6).[22]

The Coryllos Assessment is another common

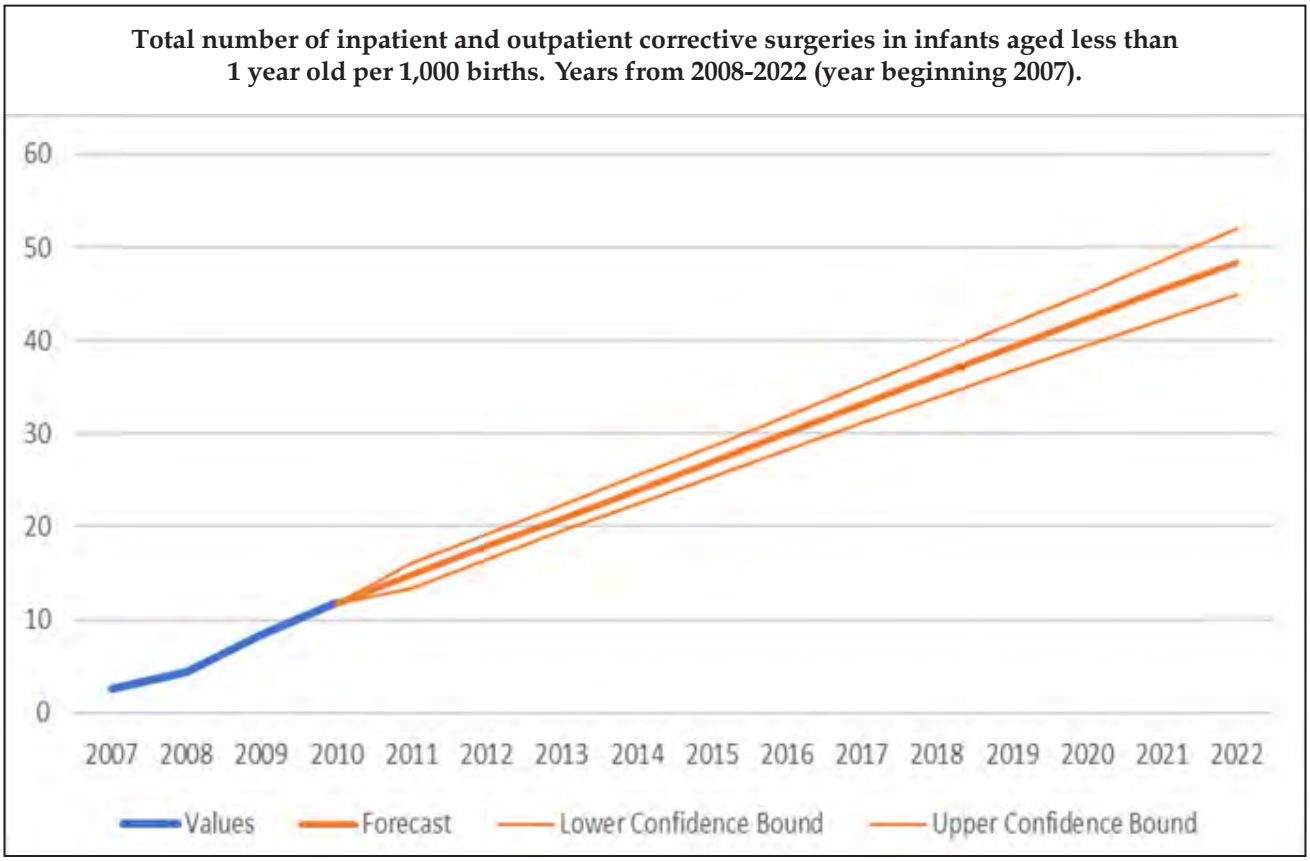


Table 4. Forecast for Trend in total number of inpatients and outpatient tongue tie (Ankyloglossia) corrective surgeries in infants aged less than 1 year old per 1000 live births at SHA's in England from the year beginning 2007.

assessment tool. This appraises the frenulum physically, observing it situated in position with the tongue and the mouth floor.[24] There are four types of classification including two types of anterior and two posterior types as seen in table 7 by Maya-Enero.[25] Type 4 is unique in that it seems to be tight lingual fascia, and an indication that the baby should be examined for related musculoskeletal issues. Amir et al,[23] reviewed the Hazelbaker assessment tool in 2006. In their study diagnostic items were condensed to improve diagnostic effectiveness.[2,23] The three items considered included tongue lateralization, lift and extension, each also showed a high inter-observer agreement with a Kappa index of 0.65.[23] Kotlow[26] also produced a tongue tie assessment tool evaluating individuals under three diagnostic criteria.[26] These include symptoms exhibited (by mother or infant) which can be associated to poor latch, appearance and function of tongue and maxillary lip, and assessment for lip tie.

The Bristol Tongue Assessment Tool (BTAT) was developed aiming to produce an objective tool to provide consistent assessment of tongue appearance and function for infants with ankyloglossia.[27] This tool scores infants from 0-2 in four different categories. The four categories are tongue tip appearance, attachment to the lower gum ridge, lift of the tongue and protrusion of the tongue. When the score is summed, totals of 0-3 are considered indicative of more severe reduction in tongue function.[27] This tool was found to have good consistency (0.760) and showed significant correlation with Hazelbaker's assessment tool (0.89) which indicates the simpler tool could be a more objective alternative.[27] Other tools do exist, but these demonstrate the variety of assessments employed by health care providers and may explain some of the inconsistencies in diagnosis.

Current Treatment

As there are various treatments available in the approach to AG care, the choice can depend on the age of the infant or child at diagnosis, the practitioner responsible for intervention, preferences of the guardian and location of the patient.[5] Patients may immediately have a LF division via frenotomy or may trial manual care to assess improvements. Lactation consult, or BF council may be approached to see if there is any advice applicable to mother and infant to improve feeding efficiency.[28] In older patients beginning to communicate, speech and language therapists (SLT) can be used as part of a multidisciplinary team to improve and aid the function of the tongue.[29]

- Latching difficulty.
- Painful and/ or cracked nipple.
- Prolonged or very frequent feeds.
- Tongue cannot extend beyond the lips.
- Stunting. (Growth).
- Recurrent mastitis.
- Callus from breast feeding. (This is formed on the breast).
- Facial asymmetry.
- Twisted mouth when open.
- Tongue remains down during crying.
- Gothic palate. (Impaired or high palate).
- Cheek hypertrophy.

Table 5. Manifestations of AG.[2]

Appearance items
Appearance of tongue when lifted
Elasticity of frenulum
Length of LF when tongue is lifted
Attachment of LF to Tongue
Attachment of LF to inferior alveolar ridge.
Functional Items
Lateralization
Tongue Lift
Tongue Extension
Spread of Anterior Tongue
Cupping
Peristalsis
Snapback

Table 6. Items of Hazelbaker Assessment Tool.[21]

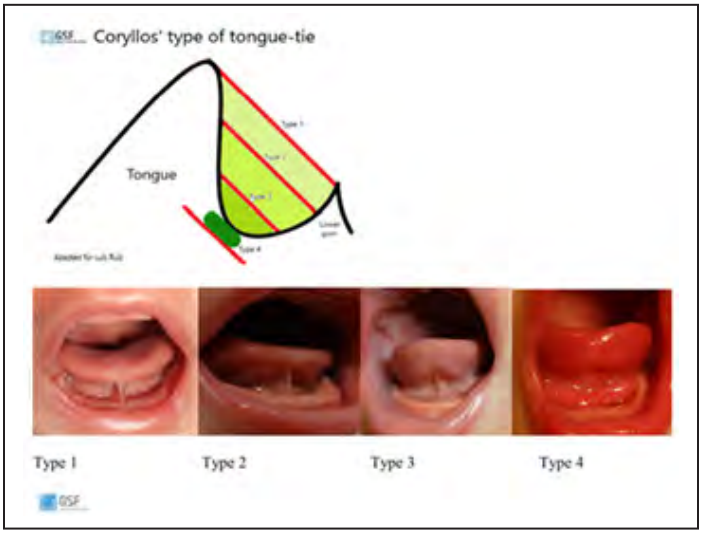


Table 7. Coryllos Classification Types (Maya-Enero, López-Vílchez, Luis Ruiz; 2021)[25] Photos by Dr. Luis Ruiz-Guzman; with permission from Dr. Silvia Maya-Enero.

Manual therapists such as chiropractors and osteopaths deploy tactile skills to assess areas of tension surrounding the jaw, cranium, neck and floor of the mouth, before soft mobilization techniques are used to decrease tissue tension.[30]

The complication rates of these interventions are hard to predict because of the lack of data recorded on the affects they have on patients with AG complaints. Despite this, Dixon et al.[31] discusses the benefits of a multifaceted approach to AG prior to frenotomy.[31] They suggest patients should receive a thorough AG diagnosis using the BTAT and lactation consult.[31] Their findings showed a significant improvement in breastfeeding rates with no difference from those who received a frenotomy.[31] A recent AG consensus carried out in Australia proposed a similar package of care in the form of a management pathway for those with AG diagnoses and feeding issues.[28] This suggested a non-surgical management and a reassessment of feeding before approaching surgical consult.[28] Following a management pathway will allow infants with feeding difficulties to trial conservative care and management under a range of medical professionals prior to seeking operative attention. Research evidence, over-all, is lacking for treatment options for AG.

Currently, surgical treatment is recommended for AG if it is associated with BF problems that are not resolving conservatively.[2] Some researchers believe, depending on the degree of restriction, that operative intervention can affect subjects' speech, oral function and oral posture.[32] However, evidence behind current surgical division is inconsistent. A systematic review by Visconti et al.[33] highlighted that although improvements are made in many surgical trials, the diagnosis and assessment vary, and progress is unpredictable.[33] Ghaheri et al[34] produced a randomized controlled trial on frenotomy for infants with diagnosed posterior tongue-ties (PTT).[34] They claimed that division both subjectively and objectively improved infant feeding. Ghaheri et al[34] found the 'within-burst' suck period of infants improved in surgical groups to -0.14 from 0.9, compared to 0.27 from 1.0 found in control groups.[34] This suggested that in a burst of sucks, the infant can produce faster sucks if the tongue is divided.[34] Using the Breastfeeding Self-Efficacy Scale Short Form (BSES-SF) they subjectively determined BF confidence scores improved from 37.5 to 50.9.[34] However, the study itself had a small sample size and there was no follow up beyond 10 days.[34]

Surgery involves the division of the LF; this is a fast procedure which can be completed in the office of a trained professional, establishing it as the most common technique performed on infants.[1,27] These professionals include dentists (DDS, DMD), oral surgeons and other medical physicians (MD's with specialties like (but not

limited to) otolaryngology, pediatric surgery, general pediatricians), physician assistants (PA) and advanced practice registered nurses (APRNs), midwives and properly trained international board of certified lactation consultants (IBCLC).[1,26,27] Depending on the regulations in their country, state or province, IBCLCs would be required to have additional credentials that qualify them to perform this procedure under their scope of practice.

The procedure is sometimes accompanied by anaesthesia (24%) in the form of sucrose both pre and post-surgery.[2] Further, a 2016 study conducted by Shavit et al[35] investigated the topical administration of some anaesthetizing agents used within frenotomy procedures, finding them ineffective (benzocaine and tetracaine).[35] Shavit et al[35] suggested that the search should continue for effective pain management for infants during frenotomy procedures.[35] After surgical intervention, the infant should be placed in skin-to-skin contact with the mother immediately to initiate feeding and assist with pain relief. Sucrose could be reapplied if necessary.[36]

When performing the frenotomy, the infant may be wrapped securely in a blanket or in a structured papoose consisting of a board, head rest and Velcro fastened wraps to effectively limit movement. To keep the mouth open, an assistant gently holds down the chin before the clinician lifts the tongue with their fingers or an appropriate tool. A small incision is then made approximately 2mm in the central mucous membrane using surgical scissors or a laser.[37] Special care is taken to avoid damage to the base of the tongue due to its vascularization and the position of the Wharton duct.[38] Kenny-Scherber and Newman[39] discuss applying pressure with the index finger on the incision area, after the initial cut, to broaden any division made.[39]

Frenectomy and frenuloplasty are different procedures that often get interpreted as frenotomy. Frenectomy involves the complete removal of the frenulum either using a scalpel or laser.[1,33] Whereas a frenuloplasty is a restructuring of the frenulum using plastic surgery to minimise any scar tissue development.[33,40] These techniques are beyond the scope of this paper.

Complications

Current rates of complications with frenotomies are relatively low ranging from 1-9% across different studies.[41,42] Despite this, there are many different complications which have been observed from this procedure. Previously frenotomies have been described as both pain and risk free. However, there is now an understanding that this is not the case, as infants can experience prolonged pain and the possible onset of other symptoms.[41,43,44] Pain is not a complication that should be overlooked in this vulnerable period of infant development. Victoria and Murphy[45] discussed that an exposure to pain in early life can result in hyper- or hyposensitive phenotype in response to short term or lasting pain and stress in later life.[45]

Walsh and Tunkel[21] reported a series of different complications and their prevalence including bleeding (3-5%), recurrence (5%), lip or Whartons duct injury, infection, injury to the lingual nerve, formation of mucous retention cyst, pain and failure of technique to improve BF.[2] Rates of oral or breast aversion are also significant enough to be documented in studies as infants negatively associate objects in their mouth with pain or stress, disrupting a crucial relationship between a mother and their infant.[46] Hale et al[41] emphasised other complications often missed in other papers reporting incidences after division.[41] They noticed infants were having delays in the diagnosis of underlying conditions, which had been overlooked in favour of treating AG.[41] One case involving a cardiac disorder, another severe dehydration and weight loss and one case involved a failure to adequately establish feeding.[41] The same paper also mentioned the complication of financial burden which often gets overlooked as there can be a high cost for the procedure.[41]

Other case reports have highlighted additional circumstances where further complications have arisen such as Ludwig Angina, two cases of severe hypovolaemic shock and two cases of frenotomies leading to airway obstructions in patients with Pierre Robin Sequence.[47-50]

Financial burdens and complications to infant not only interfere with BF but also affect a parent's ability to consent to continued BF. They could feel additional pressure to continue BF due to the possible trauma placed on the infant or the financial sacrifice made to carry out the procedure.

It is not uncommon for invasive procedures to have secondary impacts and complications. There are risks of complication in all surgical interventions such as wound dehiscence, infection, excess bleeding, nerve injury or scar and tissue adhesions, which could affect future function of the body.[51] It is important to address other types of therapies as surgical procedures are not without serious side effects. Therefore, should surgical procedures be used in any but the most egregious cases? There may be other considerations with AG that should be considered first.

A 2019 study by Mills et al[6] declared the lingual frenulum is less a band of elastic tissue waiting to be snapped back into place, but instead a fibrous fold in a layer of fascia. Fascia itself is known to have various anatomical functions such as stabilization, imparting of strength, maintaining vessel patency, separation of muscles and enclosure of different organs.[52] However, the function of LF is to stabilize the tongue under tension against resulting diverse vectors

of forces.[6] This static fascial role would suggest that the LF cannot be tight at all. It is more a fascial captive of the surrounding tissue, only to be considered taut if the attaching muscles or structures are dysfunctional. This emphasizes the concern of whether the LF should be cut at all.

It is considered in some literature, that the lingual frenulum plays part in a chain of fascia and muscle from the tongue and floor of the mouth, down the anterior neck and abdomen, via the genioglossus, then geniohyoid and then mylohyoid muscles before inserting onto the hyoid bone in the neck.[53-55] Myers[55] explored this theory further when discussing the next linked tissues. They suggested that stresses on the hyoid bones could be conceived in the anterior chest, then in the abdomen, before continuing down these myofascial and kinematic chains until reaching the ankles and feet.[55] Due to the relationship between these muscles and tissues, it is reasonable to assume that there is a chain reaction through the body.[55]

Compensatory muscular tension is a regular complication of AG. If the muscle surrounding the jaw and oral cavity becomes tight due to a restriction in tongue movement, associated muscles will also be affected.[37] These longitudinal muscular continuums still function; however, adaptations can interfere with central pattern generators located within the brain stem and spinal cord.[37,56] These important areas, responsible for the production of movements and roles governing an infant's ability to suck swallow and breathe, may now perform suboptimally.[37,56] The immediate release of these regionally affected structures could induce a fast increase to the motion in an ill prepared infant leaving them orthopedically susceptible.[37,56] A 2012 investigation by Borstad and Szucs[57] in adults, demonstrated this hypothesis in breast cancer suffers, revealing that patients who had breast surgery were consequently vulnerable to further orthopedic problems as kinematics in the shoulder had been altered.[57] These patients also scored significantly higher on pain scores and had altered recreation and active function.[57]

If restriction at the LF can affect up to the occiput and down to the abdomen and lower limbs, it is feasible that these areas can equally affect the LF causing tension in other areas of the kinematic chain.[55] Therefore, consequent tension caused by birthing trauma can follow these anatomical cascades increasing tension at the LF putting the infant at a greater risk of an AG diagnosis.[56,58] Gottlieb[59] suggested that an increase in minor birthing trauma correlates with an increase in the levels of intervention in the current birth process.[59] These interventions have musculoskeletal consequences on the infant.[58,59] Therefore, it is not irrational to hypothesize that frenotomy populations accordingly could decrease if tension from traumatic events were treated effectively.

When considering contributing factors to a blanket diagnosis of AG, the palate is a vital part of the feeding mechanism that should not be dismissed. When feeding, the tongue works in conjunction with the palate of the mouth to create a negative pressure to initiate a milk ejection reflex in the breast.[37,60] If the palate is too high, from a congenital abnormality or dysfunctional muscles such as the palatoglossus, when the infant draws the breast into the mouth, they will not be able to feed effectively.[37] This ineffective ability to suckle will likely, in individuals with a taut LF, be diagnosed as AG. However this judgement is ambiguous.[56] When treating the palate of an infant, Williams[58] suggested that the skull is a useful tool for assessment and therapy.[58] Williams looks at the maxilla, which can be affected by birthing trauma, to be a contributor to hard palate deformation.[58] This can be treated with soft internal manipulation using a touch and hold technique as advocated by Williams.[58] However, the maxilla should not be a practitioner's only therapeutic focus. Instead, look to the occiput to influence any tightness or imbalance within the sphenoid prior to attempting to treat the maxilla.[61]

Davis discusseed how the kinesiopathological pattern of an infant can be optimised by restoring typical function of the sphenobasilar mechanism.[61] Although there is insufficient research to support manual therapy alone in the treatment of AG, it is logical that considerations need to be made for all of the vital surrounding tissues in the newborn.

The relationship of the hypoglossal nerve should also be discussed as a factor in relation to AG. This nerve supplies the tongue, travelling through the hypoglossal foramen at the occipital condyle to innervate the tongue in the mouth. In neonates the occipital condyle is cartilaginous and in two separate parts. Carriero[62] discussed how unwanted distortion of these components may compromise the lumen the nerve travels through.[62] This could lead to reduction in sucking activity from irritation to the hypoglossal nerve at the level of the occipital canal.[58] Furthermore, an obstruction from biomechanical compromise, in the sensorimotor integration mediating part of the central nervous system (which informs the musculoskeletal system of environmental demands) can present as dysfunction in an infant such as ineffective feeding.[46] This draws interest to the base of the skull when thinking about treating AG. Anatomical science demonstrates how the oropharyngeal system is effectively tethered to the base of the cranium. The muscles of the tongue attach to the mandible, the temporal bone and the hyoid, while the hyoid attaches to the temporal bones via the stylohyoid ligaments and muscles and digastric.[58] Therefore, it is plausible to assume that if the range and combined movements of these associated tissues and bones are dysfunctional, the suckling and feeding of the neonate will be affected. Treating these areas of dysfunction may improve the struggling infant's ability to feed efficiently.

All procedures used to treat ankyloglossia in infants need to be properly researched to determine the best and safest evidence-based approach to improve the infant's comfort and function. Chiropractic care has been found safe using manual therapies in infant care.[63] At a minimum, all clinicians who treat these infants should keep accurate and complete records and share them across professions to help determine clinically useful treatment options.

Conclusion

Ankyloglossia is a common diagnosis in breastfeeding difficulties. Frenotomy is a quick procedure that in some cases facilitates the latching of the infant at the breast almost immediately. However, it is not without risks and lacks high-quality research evidence. There are many variables that need to be revisited before a practitioner can assess infants for ankyloglossia with absolute clarity. The perfect clinical scenario calls for a universal assessment tool and diagnosis pathway to be utilized by all clinicians, allowing practitioners to approach feeding difficulties conservatively prior to the division of the lingual frenulum. This will ensure all care is individualized and potentially harmful surgical intervention is avoided where possible.

References:

1. Junquiera MA, Cunha NN, Silva LLC, Araújo LB, Moretti AB, Couto Filho CE and Sakai VT. Surgical Techniques for the Treatment of Ankyloglossia in Children: a case series. *Journal of Application of Oral Science*. 2014;22(3):241-8.

2. Costa-Romero M, Espinola-Docio B, Paricio-Talayero JM and Diaz-Gomez NM. Ankyloglossia in breastfeeding infants. An update. *Archive of Argentinian Paediatrics*. 2021;119(6): e600-e609.

3. Hazelbaker AK, Baeza C, Watson Genna C, Murphy J, Kaplan M, Douglas P, Martinelli R, Marchesan I and Smillie C. Incidence and prevalence of tongue-tie. *Clinical Lactation*. 2017;8(3): doi:10.1891/2158-0782.8.3.89.

4. Hill RR, Lee CS and Pados BF. The prevalence of ankyloglossia in children aged <1 year: a systematic review and meta-analysis. *Pediatric research*. 2021;90(2): 259—266.

5. Department of Health and Social Care. Infant feeding profiles 2010 to 2011. Gov.uk. https://www.gov.uk/government/publications/infant-feeding-profiles-2010-to-2011. Published July 20, 2013. Accessed April 11, 2023.

6. Mills N, Pransky SM, Geddes DT and Mirjalili SA. What is a tongue tie? Defining the anatomy of the in-situ lingual frenulum. *Clin Anat*. 2019;Sep;32(6):749-761. doi:10.1002/ca.23343. Epub 2019 Feb 19. PMID: 30701608; PMCID: PMC6850428.

7. Geddes DT, Kent JC, McCellan HL, Garbin CP, Chadwick LM and Hartmann PE. Sucking Characteristics of a successfully breastfeeding infants with ankyloglossia: a case series. *Acta Paediatrica*. 2010;99(2):301-3.

8. Gonzalez JD, Costa Romero M, Riano Galan I, Gonzalez Martinez MT. Prevalencia de anquiloglosia en recien nacidos en el principado de Asturias. *An Pediatrician* (Barc).2014;81(2):115-9.

9. Bussi MT, Corrêa CC, Cassettari AJ, Giacomin LT, Faria AC, Moreira APSM, Magalhães I, Cunha MOD, Weber SAT, Zancanella E and Machado Júnior AJ. Is ankyloglossia associated with obstructive sleep apnea? *Brazilian journal of otorhinolaryngology.* 2022 88(S1); S156—S162.

10. National Institute of Health Excellence (NICE). Division of ankyloglossia (tongue-tie) for breastfeeding. *NICE Guidance*. https://www.nice.org.uk/guidance/ipg149. Published December 14, 2005. Updated January 22, 2012. Accessed April 11, 2023.

11. Lokossou GAG, Kouakanou L, Schumacher A and Zenclussen AC. Human Breast Milk: From Food to Active Immune Response with Disease Protection in Infants and Mothers. *Frontier in Immunology*. 2022; Vol 30:849012.

12. Hinde K and German JB. Food in an evolutionary context: insights from mother's milk. *Journal of Science food and agriculture*. 2012; 92: 2219-2223.

13. Walker A. Breast Milk as the Gold standard for Protective Nutrients. *Journal Paediatrics*. 2010;153:S3-7.

14. Krol KM and Grossmann T. Psychological effects of breastfeeding on children and mothers. *Bundesundheitsblatt Gesundheitsforschung Gesundheitsschutz*. 2018; 61: 977-85. doi:10.1007/s00103-018-2769.

15. Deoni S, Dean D, Joelson S, O'Regan J and Schneider N. Early nutrition influences developmental myelination and cognition in infants and young children. *Neuroimage*. 2018; 178: 649-659. doi:10.1016/j.neuroimage.2017.12.056. Epub 2017 Dec 20. PMID: 29277402; PMCID: PMC6540800.

16. LeFort Y, Evans A, Livingstone V, Douglas P, Dahlquist N, Donnelly B, Leeper K, Lappin S and Academy of Breastfeeding Medicine. Academy of Breastfeeding Medicine Position Statement on Ankyloglossia in Breastfeeding Dyads. *Breastfeeding Medicine*. 2021;16:278-281.

17. Todd DA, Hogan MJ. Tongue-tie in the newborn: early diagnosis and division prevents poor breastfeeding outcomes. *Breastfeed Rev*. 2015;23:11—16.

18. Bellù R and Condò M. Breastfeeding promotion: evidence and problems. *Medical and surgical pediatrics*. 2017.39(2),156.

19. Ip S, Chung M, Raman G, Chew P, Magula N, DeVine D, Trikalinos T and Lau J. Breastfeeding and maternal and infant health outcomes in developed countries. *Evidence report/technology assessment*. 2007;153:1—186.

20. Gonzalez JD, Costa Romero M, Riano Galan I, Gonzalez Martinez MT. Prevalencia de anquiloglosia en recien nacidos en el principado de Asturias. *An Pediatrician* (Barc). 2014;81(2):115-9.

21. Walsh J and Tunkel D. Diagnosis and treatment of ankyloglossia in newborns and infants. A review. *J A M A Otolaryngeal Head Neck Surgery*. 2017;143(10):1032-9.

22. Hazelbaker A. The Assessment tool for lingual frenulum function (ATLFF): use in lactation consultant private practise [Dissertation]. Pasadena, CA; Pacific Oaks College: 1993.

23. Amir LH, James JP and Donath SM. Reliability of the Hazelbaker assessment tool for lingual frenulum function. *International Breastfeeding Journal*. 2006;1(1):3.

24: Coryllos E, Watson Genna C and Salloum AC. Congenital tongue-tie and its impact on breastfeeding. *Breastfeeding best for infant and mother.* 2004;Summer:1-11.

25. Maya-Enero S, Perez-Perez M, Ruiz-Guzman L, Duran-Jorda X and Lopez-Vilchez MA. Prevalance of neonatal ankyloglossia in a tertiary care hospital in spain: a transverse cross-sectional study. *European journal of Paediatrics*. 2020. [Online] doi.org/10.1007/s00431-020-03781-7 [Accessed 21.10.2022].

26. Kotlow L. TOTS —Tethered Oral Tissues The Assessment and Diagnosis of the Tongue and Upper Lip Ties in Breastfeeding. *Oralhealth*. 2015; Mar: 64-70.

27. Ingram J, Johnson D, Copeland M, Churchill C, Taylor H, Emond A. The development of a tongue assessment tool to assist with tongue-tie identification. *Arch Dis Child Fetal Neonatal Ed*. 2015;100(4):344-8.

28. Australian Dental Association. Ankyloglossia and Oral Frena Consensus Statement [Online]. *NSW:Australian Dental Association*; 2020.

29. Caloway C, Hersh CJ, Baars R, Sally S, Diericks G and Hartnick CJ. Association of Feeding Evaluation With Frenotomy Rates in Infants With Breastfeeding Difficulties. *J A M A Otolaryngology—Head & Neck Surgery*. 2019;145(9):817-822.

30. Dorough A and Vallone S. Differentiating the impact of biomechanical forces of labor and deliver vs. the effect of a posterior tongue tie on neonatal and infant feeding dysfunction: a clinical evaluation. *Journal of Clinical Chiropractic Paediatrics*. 2022;21(2):1893-1901.

31. Dixon B, Gray J, Elliot N, Shand B and Lynn A. A multifaceted programme to reduce the rate of tongue-tie release surgery in newborn infants: Observational study. *International Journal of Pediatric Otorhinolaryngology*. 2018;113:156-163.

32. Marchesan IQ, Martinelli RL and Gusmão RJ. Lingual frenulum: changes after frenectomy. *J Soc Bras Fonoaudiol*. 2012;24(4):409-12.

33. Visconti A, Hayes E, Ealy K and Scarborough DR. A systematic review: The effects of frenotomy on breastfeeding and speech in children with ankyloglossia. *International Journal of Speech-Language Pathology*. 2021;23:349-358

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

35. Shavit I, Peri-Front Y, Rosen-Walther A, Grunau RE, Neuman G, Nachmani O, Koren G and Aizenbud D. A Randomized Trial to Evaluate the Effect of Two Topical Anesthetics on Pain Response During Frenotomy in Young Infants. *Pain medicine*. 2017 18(2), 356—362.

36. Efe E and Ozer ZC. The use of breast-feeding for pain relief during neonatal immunization injections. *Applied nursing research: ANR*, 2007;20(1):10-16.

37. Watson Genna C. Supporting Sucking skills in breastfeeding infants. Burlington Massachusetts: Jones and Bartlett Learning; 2017.

38. Cuestas G, Demarchi V, Martinez Corvalan MP, Razetti J and Boccio C. Tratamiento quirugico del frenillo lingual corto en ninos. *Arch argent pediatr*. 2014;112(6):567-70.

39. Kenny-Scherber AC and Newman J. Office base-frenotomoy for ankyloglossia and problematic breastfeeding. *Can Fam Physician*. 2016;62(7):570-1.

40. Baker AR and Car MM. Surgical treatment of ankyloglossia. *Operative Techniques in Otolaryngology.* 2015;26:28—32

41. Hale M, Mills N, Edmonds L, Dawes P, Dickson N, Barker D and Wheeler BJ. Complications following frenotomy for ankyloglossia: A 24-month prospective New Zealand Paediatric Surveillance Unit study. *Journal of Paediatrics and Child Health*. 2019;56(4):557—562.

42. Cuestas G, Demarchi V, Martinez Corvalan MP, Razetti J and Boccio C. Tratamiento quirugico del frenillo lingual corto en ninos. *Arch argent pediatr. 2014;112(6):567-70.*

43. Levkovich R, Sidebotham M, Vaughan K and Dietsch E. Ankyloglossia (Tongue-Tie)–To Snip or Not to Snip: An Integrative Literature Review. *International Journal of Childbirth*. 2017;7(3):126-138.

44. DiLorenzo M, Riddell RP and Holsti L. Beyond Acute Pain: Understanding Chronic Pain in Infancy. *Children*. 2016;3(4):26.

45. Victoria NC and Murphy AZ. Exposure to Early Life Pain: Long Term Consequences and Contributing Mechanisms. *Current opinion in behavioral sciences*. 2016;7:61—68.

46. O'Connor ME, Gilliland AM and LeFort Y. Complications and misdiagnoses associated with infant frenotomy: results of a healthcare professional survey. *Int Breastfeed J*. 2022;17(1):39.

47. Lin HW, O'Neill A, Rahbar R and Skinner ML. Ludwig's angina following frenuloplasty in an adolescent. *Int J Pediatr Otorhinolaryngol*. 2017;73(9):1313-1315.

48. Opara PI, Gabriel-Job N and Opara KO. Neonates presenting with severe complications of frenotomy: a case series. *J Med Case Rep*. 2012;6(1):77.

49. Genther DJ, Skinner ML, Bailey PJ, Capone RB and Byrne PJ. Airway obstruction after lingual frenulectomy in two infants with Pierre-Robin Sequence. *Int J Pediatr Otorhinolaryngol*. 2015;79(9):1592-1594.

50. Walsh F and Kelly D. Partial airway obstruction after lingual frenotomy. *Anesth Analg*. 1995;80(5):1066-1067.

51. Kolodziej L, Bohatyrewicz A, Kromuszczynska J, Jezierski J and Biedron M. Efficacy and complications of open and minimally invasive surgery in acute Achilles tendon rupture: a prospective randomised clinical study--preliminary report. *Int Orthop*. 2013;Apr;37(4):625-9.

52. Gatt A, Agarwal S and Zito PM. 2022. Anatomy, Fascia Layers. [Updated 2021 Jul 26]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; Jan-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK526038/.

53. McCausland, T. and Bordoni, B. 2022. Anatomy, Head and Neck, Genioglossus Muscle. [Updated 2022 Jun 11]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; Jan-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK545141/?report=classic.

54. Muscolino JE. The muscular system manual the skeletal muscles of the human body. 3rd edition. Missouri, Maryland Heights: Mosby Elsevier; 2010.

55. Myers TW. Anatomy trains myofascial meridians for manual and movement therapists. 3rd edition. England, London: Churchstone Livingstone Elsevier; 2014.

56. Hazelbaker AK. Tongue-tie morphogenesis, impact, assessment and treatment. United States of America, Ohio: Aidan and Eva Press; 2010.

57. Borstad JD and Szucs KA. Three-dimensional scapula kinematics and shoulder function examined before and after surgical treatment for breast cancer. *Human movement science*. 2012;31(2):408-18.

58. Williams SP. Pregnancy and paediatrics a chiropractic approach. England, Southhampton: Williams *Paediatrics*; 2005.

59. Gottlieb MS. Neglected spinal cord, brainstem and musculoskeletal injuries stemming from birth trauma. *J manipulative physiol ther*. 1993:16;537-3.

60. Geddes DT, Kent JC, Mitoulas LR and Hartmann PE. Tongue movement and intraoral vacuum in breastfeeding infants. *Early human development*. 2008;84:471-477.

61. Davis NJ. Chiropractic Paediatrics. 2nd Edition. Elsevier. London, United Kingdom: Churchill and Livingstone; 2010.

62. Carreiro JE. An osteopathic approach to children. Scotland, Edinburgh: Churchill Livingstone; 2003.

63. Miller J. Evidence based chiropractic care for infants rational, therapies, and outcomes. United States of America, Texas: Praeclarus Press; 2019.

Complexity of infantile colic presentations and the impact on chiropractic outcomes: A narrative review

Ramneek S. Bhogal, DC, DABCI,[1] Danielle Perna-Amari, DC, MS,[2] Stephanie O'Neill Bhogal, DC, DICCP[3]

[1]Associate Dean & Professor, Life University College of Chiropractic, Marietta, GA
[2]Private Practice, West Chester, PA
[3]Professor, Life University College of Chiropractic, Marietta, GA

Article being reviewed
Holm LV, Jarbøl DE, Christensen HW, Søndergaard J, Hestbæk L. The effect of chiropractic care on infantile colic: Results from a single-blind randomised controlled trial. *Chiropractic & Manual Therapies*. 2021;29(1). doi:10.1186/s12998-021-00371-8.

Study Objective
Review the outcomes of a single-blind RCT of chiropractic care for the treatment of colic and provide additional clinical considerations.

Study Design
Single-blind, randomized study performed at four Danish chiropractic clinics.

Study Participants
200 children were recruited, of which 185 participated in the study trial. 96 patients were randomized to the treatment group and 89 were randomized to the control arm. Children were 2 to 14 weeks of age with colic. Colic was defined as having excessive crying or fussiness for at least three hours a day for three or more days a week during the previous two weeks, in an otherwise healthy child with normal weight gain. Randomization (1:1) into the control or treatment group was directed by computer-generated allocations, stratified by age at enrollment (2-6 weeks, 7-10 weeks, or 11-14 weeks) and the treating chiropractor. Children were excluded if they had previously received chiropractic care. No ancillary treatment for colic was permitted during the study.

Study Parameters Assessed
Children participating in the treatment cohort of the study, received chiropractic care twice a week for two weeks, for a duration of five minutes per session. If they were in the control arm of the study, they were entertained for a comparable duration without receiving care. Parents kept 24-hour diaries (divided into 15-minute increments) during the two-week study to assess their child's behavior. They tracked the frequency of inconsolable crying, time the child needed to be held or rocked to limit crying, the time the child was awake and content, time spent sleeping, feeding patterns, and bowel movements. Parents were asked to record in their 24-hour diary for one to four more days after the fourth chiropractic visit, in addition to completing a final questionnaire.

Primary Outcome Measures
The primary outcome was the change in the duration of crying, with a reduction of at least one hour a day being considered clinically significant. Secondary outcomes were sleep duration, hours spent awake and content, number of bowel movements, burps, hiccups, regurgitation, satisfaction of participation in care, and status of colic.

Key Findings
The treatment group experienced an average reduction of crying by one and one-half hours, while the control group experienced a reduction by only one hour, with the change in hours of crying ranging from -8.5 to +3.5 hours. Improvement of one or more hours in crying was achieved in 63% in the treatment group and 47% in the control group. The difference between the two groups was not statistically significant when adjusted for the baseline hours of crying, age, and chiropractic clinic. Secondary outcomes were insignificantly better in the treatment group for hours of sleep and time awake and content, with no difference between groups in colic status, satisfaction, and GI symptoms. More than 90% of parents were satisfied with participation.

Practice Implications and Discussion
In the ever-evolving landscape of understanding colic in infants, significant dialogue has turned to the impact chiropractic care has on this presentation. Pediatric chiropractic care is a growing specialization within the chiropractic profession with a multitude of accredited institutions and private organizations providing education and post-graduate training. A survey of the profession indicated that, on average, 17% of chiropractic patients were under 17 years of age, while pediatric-trained chiropractors had 38.7% of their patient base in the same age demographic.[1] Clearly, parents are seeking chiropractic care for infants and are becoming more informed about the impact care has on pediatric health issues, inclusive of colic. From a clinical awareness perspective, the dialogue must shift toward effectiveness of chiropractic care for those with colic and consider other clinical or lifestyle factors that may impact its efficacy.

The investigators in this study looked to establish if chiropractic care was effective in cases of infantile colic and were guided by several previous studies suggesting that it was.[2-8] They emphasized that, unlike previous studies,

this study was unique with a well-powered design that included both parental blinding and a larger cohort.

With the treatment arm of this randomized clinical trial seeing a reduction of colic-related crying by an average of one and one-half hours juxtaposed with the control arm seeing a one hour reduction, the study suggested there was a small positive effect of chiropractic care on infantile colic, but the clinical significance was debatable because it was not statistically significant. It is important to note that the study also concluded that while the mean difference between the groups was small, large individual differences were noted which contributed to investigating subgroups of children. This highlighted an opportunity for further discussion.

While not clearly stated in the original study, statements made by the authors in the conclusion open the clinical discussion and consideration of various factors that could impact efficacy. One of the most critical areas of discussion is the method of intervention, specifically, how the biomechanical dysfunction was analyzed. The study focused on musculoskeletal methodology inclusive of visible and palpable asymmetries, motion restriction, and areas of tenderness. Given the current understanding of the vertebral subluxation complex being inclusive of a neuropathology, no clear assessment was utilized in obtaining a neurological manifestation of biomechanical dysfunction.[9] This could involve a variety of dermal thermographic findings, and/or sudoriferous changes in the skin. While the attention given to the musculoskeletal component of biomechanical dysfunction is noteworthy, a more thorough analysis, inclusive of a functional neurological finding could certainly impact efficacy.[10] Lending support to this position is the use of an objective measure such as heart rate variability, which has demonstrated that there can be significant improvements in visceral neurologic function, an indirect measure of autonomic nervous system function, with the use of manipulative therapies.[11] Clinical practitioners will posit that their improved patient outcomes are proof of this concept and that neurologically informed procedures, as part of an analysis, need to be given consideration.

A second area of discussion in the study, that could certainly impact efficacy, is the singularity of adjusting technique utilized. "Very light short term-pressure with fingertips" was certainly an acceptable approach to chiropractic care, however, the reviewing authors agree that it comes with its limitations. This is especially true if the motion dysfunction of the subluxation is remarkable and leads to a significant fixation and resulting joint misalignment. Approaches such as cranial/dural, instrument assisted, drop-mechanism, and pediatric modified high velocity low amplitude adjusting are also safely utilized techniques by pediatric chiropractors. Clinical observations and case studies suggested that the use of these techniques is correlated with positive outcomes in pediatric care and should be incorporated in the dialogue of improving outcomes with infantile colic.[2-8]

Another area of this study that warrants attention is the duration of care, which can significantly impact outcomes. The study commented on the project period being two weeks with four chiropractic visits, and how this revolved around the parental willingness to accept the study. The authors of this review acknowledge that parents often are navigating health care within the confines of time and money, and this certainly can impact healthcare access. Pediatric chiropractors must strike a balance between setting realistic expectations and management of parental resources. Studies for spinal manipulation and cranial therapies for infants with colic typically lasted 2-4 weeks in duration, with an average total of four visits. While most of these studies showed an initial improvement in outcomes with chiropractic care, this could also mean that these initial investigatory durations may not be enough time to determine dose-dependent trends or to determine what is a realistic course of care to support an infant with colic. This must then be reconciled with pediatric practitioners who value care beyond the acute presentation phase for the maintenance of health and function.

While the pathogenesis of colic is still not well understood, associations are made with a variety of factors ranging from gastrointestinal status, gestational and parturition distress, to biomechanical dysfunction. The developing fetal microbiome changes in response to the maternal microbiota, delivery mode, how the infant is fed, pharmaceutical and environmental exposure including the members of the family and family pets.[12] Interestingly, in this study, the baseline characteristics of families and infants revealed that 21 study participants had a planned cesarean birth, for example, of which, 15 were in the treatment arm as opposed to seven in the control arm. While a recent study had concluded that exposure to the maternal vaginal microbiome during childbirth did not impact the development of the infant gut biome,[13] opposing perspectives do exist. Two previous studies supported that delivery modes can impact the infant gut biome wherein cesarean born infants tend to have less intrinsically diverse gut microbiota.[14,15] A further positive correlation existed with the restoration of microbiota and crying time reduction.[16] With this rationale in hand, the reviewing authors put forward that having double the number of potentially dysbiotic participants as a result of mode of delivery in the treatment arm, likely detracted from the positive outcome of reduced crying time.

In further inquiry into participant allocation, 50 participants reported that there was a severe incident in the family during pregnancy. Of these 50 participants, 31 were in the treatment group as opposed to 19 in the control group. A

recent systematic review and meta-analysis established a relationship between greater maternal stress and the development of disease, inclusive of infantile colic.[17] Once again, given the increased number of stressed participants in the treatment arm, it is plausible that the reduction in crying may be more significant than the statistical analysis recognized.

In closing, it is imperative that clinicians recognize that various areas of pathogenesis may be at play in infantile colic. These need to be reconciled with clinical outcomes when considering the implementation of chiropractic care as an intervention for infantile colic. Overall, the studies show a cumulative range of two hours to ≥ seven hours in the reduction in crying per week in infants with colic who received care. With adverse events being rare and avoidable with proper examination and adjustments modified for pediatric patients, manual therapies such as chiropractic are a low-risk care method worthy of consideration for infants with colic. Ancillary lifestyle factors contributing to improved gut health and maternal stress reduction are also noteworthy when considering care plans. With all these factors at the forefront, studies with more comprehensive design are needed to fortify these positions further.

Conflicts to disclose
The completion of this review was not dependent upon any external funding from any entity and the authors have no conflicts of interest to disclose.

References:

1. Pohlman K, Hondras M, Long C, Haan A. Practice patterns of doctors of chiropractic with pediatric diplomate: a cross sectional survey. *BMC Complement Altern Med*. 2010;10:26.

2. Browning M, Miller J. Comparison of the short-term effects of chiropractic spinal manipulation and occipito-sacral decompression in the treatment of infant colic: a single-blinded, randomized, comparison trial. *Clin Chiropr*. 2008;11(3):122—9.

3. Buffone F, Monacis D, Tarantino AG, Dal Farra F, Bergna A, Agosti M, Vismara L. Osteopathic Treatment for Gastrointestinal Disorders in Term and Preterm Infants: A Systematic Review and Meta-Analysis. *Healthcare*. 2022; 10(8):1525.

4. Dobson D, Lucassen PL, Miller JJ, Vlieger AM, Prescott P, Lewith G. Manipulative therapies for infantile colic. Cochrane Database Syst Rev. 2012;12:CD004796. Published 2012 Dec 12. doi:10.1002/14651858.CD004796.pub2.

5. Castejón-Castejón M, Murcia-González MA, Martínez Gil JL, et al. Effectiveness of craniosacral therapy in the treatment of infantile colic. A randomized controlled trial. *Complementary Therapies in Medicine*. 2019;47:102164. doi:10.1016/j.ctim.2019.07.023.

6. Hayden C, Mullinger B. A preliminary assessment of the impact of cranial osteopathy for the relief of infantile colic. *Complement Ther Clin Pract*. 2006;12(2):83-90. doi:10.1016/j.ctcp.2005.12.005.

7. Miller JE, Phillips HL. Long-term effects of infant colic: a survey comparison of chiropractic treatment and nontreatment groups. *J Manipulative Physiol Ther*. 2009;32(8):635-638. doi:10.1016/j.jmpt.2009.08.017.

8. Miller JE, Newell D, Bolton JE. Efficacy of chiropractic manual therapy on infant colic: a pragmatic single-blind, randomized controlled trial. *J Manipulative Physiol Ther*. 2012;35(8):600-607. doi:10.1016/j.jmpt.2012.09.010.

9. Senzon SA. The Chiropractic Vertebral Subluxation Part 1: Introduction. *J Chiropr Humanit*. 2019;25:10-21. Published 2019 Apr 6. doi:10.1016/j.echu.2018.10.002.

10. Owens EF Jr, Hart JF, Donofrio JJ, Haralambous J, Mierzejewski E. Paraspinal skin temperature patterns: an interexaminer and intraexaminer reliability study. *J Manipulative Physiol Ther*. 2004;27(3):155-159. doi.org/10.1016/j.jmpt.2003.12.019.

11. Ruffini N, D'Alessandro G, Mariani N, Pollastrelli A, Cardinali L, Cerritelli F. Variations of high frequency parameter of heart rate variability following osteopathic manipulative treatment in healthy subjects compared to control group and sham therapy: randomized controlled trial. *Front Neurosci*. 2015;9:272. doi:10.3389/fnins.2015.00272.

12. Jeong S. Factors influencing development of the infant microbiota: from prenatal period to early infancy. *Clin Exp Pediatr*. 2022;65(9):439-447. doi:10.3345/cep.2021.00955.

13. Dos Santos SJ, Pakzad Z, Albert AYK, et al. Maternal vaginal microbiome composition does not affect development of the infant gut microbiome in early life. *Front Cell Infect Microbiol*. 2023;13:1144254. Published 2023 Mar 30. doi:10.3389/fcimb.2023.1144254.

14. Zhang C, Li L, Jin B, et al. The Effects of Delivery Mode on the Gut Microbiota and Health: State of Art. *Front Microbiol*. 2021;12:724449. Published 2021 Dec 23. doi:10.3389/fmicb.2021.724449.

15. Mueller NT, Differding MK, Østbye T, Hoyo C, Benjamin-Neelon SE. Association of birth mode of delivery with infant faecal microbiota, potential pathobionts, and short chain fatty acids: a longitudinal study over the first year of life. *BJOG*. 2021;128(8):1293-1303. doi:10.1111/1471-0528.16633.

16. Nocerino R, De Filippis F, Cecere G, et al. The therapeutic efficacy of Bifidobacterium animalis subsp. lactis BB-12® in infant colic: A randomised, double blind, placebo-controlled trial. *Aliment Pharmacol Ther*. 2020;51(1):110-120. doi:10.1111/apt.15561.

17. Caparros-Gonzalez RA, Torre-Luque A, Romero-Gonzalez B, Quesada-Soto JM, Alderdice F, Peralta-Ramírez MI. Stress During Pregnancy and the Development of Diseases in the offspring: A Systematic-Review and Meta-Analysis. *Midwifery*. 2021;97:102939. doi:10.1016/j.midw.2021.102939.

JOURNAL ABSTRACTS

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

Comparison of Forces Exerted by a Chiropractor on Children and Adults During High-Speed, Low-Amplitude Spinal Manipulations: A Feasibility Study

Walter Herzog PhD; Peter Kevorkian DC; Brent Russell DC; Joel Alcantara DC

Journal of Manipulative and Physiological Therapeutics Volume 45, Issue 6, July—August 2022, Pages 389-399
doi.org/10.1016/j.jmpt.2022.09.003.

Abstract

Objective: The aim of this study was to demonstrate that quantification of the forces exerted by a single chiropractor on children and adults during high-velocity, low-amplitude spinal manipulations and the correlation of forces to age was feasible. **Methods:** The force-time profiles of high-velocity, low-amplitude spinal manipulations were measured in 48 children (109 manipulations) ranging from 14 weeks to 17 years of age, and 20 adults (49 manipulations) in a clinical setting. The measurements were taken using a thin, flexible pressure pad. Outcome variables (peak forces, preload forces, thrust forces, thrust durations, rates of force application, and thrust impulses) were quantified and compared across age groups using Kruskal-Wallis testing with Dunn post hoc analysis. Outcome variables were fitted with best-fitting linear regressions with age as the dependent variable. The level of significance for all statistical tests was set a priori at a = 0.05. **Results:** Most outcome variables increased with the age of the patient. Specifically, peak forces, thrust forces, and the rate of force application were positively correlated with age, while thrust durations remained constant across all ages and preload forces decreased slightly with patient age for cervical spine manipulations. **Conclusion:** For this single chiropractor in private practice, the forces he used increased with the age of the patient, and he thus used lower forces in children than adults. This study shows that measuring the forces used by a chiropractor in clinical practice on patients with a range of ages was feasible.

Key Indexing Terms: Manipulation, Chiropractic, Infant, Child, Manipulation, Spinal, Mechanical Phenomena.

Association Between Pregnancy-Related Hormones and Lumbopelvic Pain Characteristics in Pregnant Women: A Scoping Review

Catherine Daneau; Marième Houle; Mégane Pasquier; Stephanie-May Ruchat; Martin Descarreaux

J Manipulative Physiol Ther. 2021 Sep;44(7):573-583.
doi:10.1016/j.jmpt.2021.10.001. Epub 2021 Dec 9.

Abstract

Objectives: The objectives of this scoping review were (1) to document and quantify the potential associations between lumbopelvic pain characteristics and pregnancy-related hormones, and (2) to identify research approaches and assessment tools used to investigate lumbopelvic pain characteristics and pregnancy-related hormones. **Methods:** The literature search was conducted in six databases (MEDLINE, Academic Search Complete, Cumulative Index to Nursing and Allied Health Literature, SportDiscus, PsycINFO, and Cochrane) from inception up to March 2020 and completed using search terms relevant to pregnant women, pregnancy-related hormones, and lumbopelvic pain. The risk of bias was assessed using the characteristics recommended by Guyatt et al. for observational studies. Results: The search yielded 1015 publications from which nine met the inclusion criteria. Relaxin was the most studied pregnancy-related hormone. An association between relaxin levels and lumbopelvic pain presence or severity was found in 4 studies, while five studies did not report an association between them. One study reported an association between relaxin and lumbopelvic pain presence or severity while two studies reported no association and were considered as having a low risk of bias. One study reported measures of estrogen and progesterone levels. It showed that progesterone levels were found to be significantly higher in pregnant women with lumbopelvic pain compared to those without, while estrogen concentrations were similar in both groups. **Conclusion:** The literature showed conflicting evidence regarding the association between pregnancy-related hormones and lumbopelvic pain characteristics in pregnant women. The assessment tools used to investigate lumbopelvic pain characteristics and pregnancy-related hormones are heterogeneous across studies. Based on limited and conflicting evidence, and due to the heterogeneity of assessment tools and overall poor quality of the literature, the association between pregnancy-related hormones and lumbopelvic pain characteristics is unclear.

Keywords: Estrogen; Low Back Pain; Pelvic Girdle Pain; Pregnancy; Progesterone; Relaxin.

Adverse infant outcomes among women with sleep apnea or insomnia during pregnancy: A retrospective cohort study

Jennifer N Felder; Rebecca J Baer; Larry Rand; Kelli K Ryckman; Laura Jelliffe-Pawlowski; Aric A Prather

Sleep Health 2023 Feb;9(1):26-32.
doi:10.1016/j.sleh.2022.09.012. Epub 2022 Nov 10.

Abstract

Objective: To evaluate whether sleep apnea or insomnia among pregnant people is associated with increased risk for adverse infant outcomes. **Design:** Retrospective cohort study SETTING: California PARTICIPANTS: The sample included singleton live births. Sleep apnea and insomnia were defined based on ICD-9 and -10 codes. A referent group was selected using exact propensity score matching on maternal characteristics, obstetric factors, and infant factors among individuals without a sleep disorder. **Measurements:** Adverse infant outcomes were obtained from birth certificate, hospital discharge, and death records (eg, Apgar scores, neonatal intensive care unit (NICU) stay, infant death, long birth stay, etc.). Logistic regression was used to calculate odds of an adverse infant outcome by sleep disorder type. **Results:** Propensity-score matched controls were identified for 69.9% of the 3371 sleep apnea cases and 68.8% of the 3213 insomnia cases. Compared to the propensity-matched referent group, individuals with a diagnosis of sleep apnea (n = 2357) had infants who were more likely to have any adverse outcome, low 1-min Apgar scores, NICU stay, and an emergency room visit in the first year of life. Infants born to mothers with a diagnosis of insomnia (n = 2212) were at increased risk of few negative outcomes relative to the propensity matched referent group, with the exception of an emergency room visit. **Conclusions:** In unadjusted analyses, infants born to individuals with a diagnosis of sleep apnea or insomnia were at increased risk of several adverse outcomes. These were attenuated when using propensity score matching, suggesting these associations were driven by other comorbidities.

Keywords: Infant outcomes; Insomnia; Pregnancy; Sleep apnea.

Effect of Vitamin C Supplementation for Pregnant Smokers on Offspring Airway Function and Wheeze at Age 5 Years: Follow-up of a Randomized Clinical Trial.

Cindy T. McEvoy, MD, MCR; Lyndsey E. Shorey-Kendrick, PhD; Kristin Milner, BA1; et al.

JAMA Pediatr. 2023;177(1):16-24. doi:10.1001/jamapediatrics.2022.4401.

Abstract

Importance: Vitamin C supplementation (500 mg/d) for pregnant smokers has been reported to increase offspring airway function as measured by forced expiratory flow (FEF) through age 12 months; however, its effects on airway function at age five years remain to be assessed. **Objective:** To assess whether vitamin C supplementation in pregnant smokers is associated with increased and/or improved airway function in their offspring at age five years and whether vitamin C decreases the occurrence of wheeze. **Design, Setting, and Participants:** This study followed up the Vitamin C to Decrease the Effects of Smoking in Pregnancy on Infant Lung Function (VCSIP) double-blind, placebo-controlled randomized clinical trial conducted at three centers in the US (in Oregon, Washington, and Indiana) between 2012 and 2016. Investigators and participants remain unaware of the treatment assignments. Forced expiratory flow measurements at age five years were completed from 2018 to 2021. **Interventions:** Pregnant smokers were randomized to vitamin C (500 mg/d) or placebo treatment. **Main Outcomes and Measures:** The primary outcome was the prespecified measurement of FEF between 25% and 75% expired volume (FEF25-75) by spirometry at age five years. Secondary outcomes included FEF measurements at 50% and 75% of expiration (FEF50and FEF75), forced expiratory volume in one second (FEV1), and occurrence of wheeze. **Results:** Of the 251 pregnant smokers included in this study, 125 (49.8%) were randomized to vitamin C and 126 (50.2%) were randomized to placebo. Of 213 children from the VCSIP trial who were reconsented into this follow-up study, 192 (90.1%) had successful FEF measurements at age five years; 212 (99.5%) were included in the analysis of wheeze. Analysis of covariance demonstrated that offspring of pregnant smokers allocated to vitamin C compared with placebo had 17.2% significantly higher mean (SE) measurements of FEF25-75at age five years (1.45 [0.04] vs 1.24 [0.04] L/s; adjusted mean difference, 0.21 [95% CI, 0.13-0.30]; P <.001). Mean (SE) measurements were also significantly increased by 14.1% for FEF50(1.59 [0.04] vs 1.39 [0.04] L/s; adjusted mean difference, 0.20 [95% CI, 0.11-0.30]; P <.001), 25.9% for FEF75(0.79 [0.02] vs 0.63 [0.02] L/s; 0.16 [95% CI, 0.11-0.22]; P <.001), and 4.4% for FEV1(1.13 [0.02] vs 1.09 [0.02] L; 0.05 [95% CI, 0.01-0.09]; P =.02). In addition, offspring of pregnant smokers randomized to vitamin C had significantly decreased wheeze (28.3% vs 47.2%; estimated odds ratio, 0.41 [95% CI, 0.23-0.74]; P =.003). **Conclusions and Relevance:** In this follow-up study of offspring of pregnant smokers randomized to vitamin C vs placebo, vitamin C supplementation during pregnancy resulted in significantly increased airway function of offspring at age five years and significantly decreased the occurrence of wheeze. These findings suggest that vitamin C supplementation for pregnant smokers may decrease the effects of smoking in pregnancy on childhood airway function and respiratory health.

Trial Registration: ClinicalTrials.gov Identifier: NCT03203603.

JOURNAL ABSTRACTS

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

Treatment of infant colic with craniosacral therapy. A randomized controlled trial

Mercedes Castejón-Castejón; M A Murcia-González; J Todri; O Lena; R Chillón-Martínez

Complement Ther Med. 2022 Dec;71:102885.
doi:10.1016/j.ctim.2022.102885. Epub 2022 Sep 13

Abstract

Objective: To evaluate the number of craniosacral therapy sessions that can be helpful to obtain a resolution of the symptoms of infantile colic and to observe if there are any differences in the evolution obtained by the groups that received a different number of Craniosacral Therapy sessions at 24 days of treatment, compared with the control group which did not received any treatment. **Methods:** Fifty-eight infants with colic were randomized into two groups of which 29 babies in the control group received no treatment and those in the experimental group received 1-3 sessions of craniosacral therapy (CST) until symptoms were resolved. Evaluations were performed until day 24 of the study. In this study crying hours served as primary outcome. The secondary outcome were the hours of sleep and the severity, measured by an Infantile Colic Severity Questionnaire (ICSQ). **Results:** Significant statistical differences were observed in favor of experimental group compared to the control group on day 24 in crying hours (mean difference = 2.94, at 95 %CI = 2.30-3.58; $p < 0.001$) primary outcome, and also in hours of sleep (mean difference = 2.80; at 95 %CI = - 3.85 to - 1.73; $p < 0.001$) and colic severity (mean difference = 17.24; at 95 %CI = 14.42-20.05; $p < 0.001$) secondary outcomes. Also, the differences between the groups ≤ 2 CST sessions (n = 19), 3 CST sessions (n = 10) and control (n = 25) were statistically significant on day 24 of the treatment for crying, sleep and colic severity outcomes ($p < 0.001$). **Conclusion:** Babies with infantile colic may obtain a complete resolution of symptoms on day 24 by receiving 2 or 3 CST sessions compared to the control group, which did not receive any treatment.

Keywords: Craniosacral therapy; Crying; Infantile colic; Manual therapy; Osteopathy; Sleep.

Longitudinal Associations Between Use of Mobile Devices for Calming and Emotional Reactivity and Executive Functioning in Children Aged 3 to 5 Years

Jenny S Radesky; Niko Kaciroti; Heidi M Weeks; Alexandria Schaller; Alison L Miller

JAMA Pediatr. 2023 Jan 1;177(1):62-70.
doi:10.1001/jamapediatrics.2022.4793.

Abstract

Importance: Mobile devices are often used to keep young children occupied or calm, but it is not known whether this practice influences child development. **Objective:** To examine the longitudinal, bidirectional associations between the parent-reported frequency of using mobile devices to calm young children and children's executive functioning (EF) and emotional reactivity, testing moderation by child sex and temperament. **Design, setting, and participants:** This prospective cohort study included a community-based convenience sample of English-speaking parents of typically developing children aged three to five years. The study duration was from August 2018 to January 2020, with baseline (T1), 3-month follow-up (T2), and 6-month follow-up (T3) waves. **Exposures:** Parent-reported frequency of use of mobile devices to calm children when upset (5-point Likert scale). **Main outcomes and measures:** At each wave, the child's EF was assessed with the Behavior Rating Inventory of Executive Function-Preschool Version Global Executive Composite and emotional reactivity with the Child Behavior Checklist Emotional Reactivity subscale. Structural equation models were built to examine cross-lagged associations of the use of devices for calming, EF, and emotional reactivity, testing for moderation by child sex or temperament (Child Behavior Questionnaire-Very Short Form surgency score, median split). **Results:** Of 422 eligible parents with data at T1, 375 (88.9%) provided data at T2 and 366 (86.7%) at T3. At baseline, the mean (SD) age of the 422 children was 3.8 (0.5) years, the number of boys in the sample was 224 (53.1%), the number of individuals of non-Hispanic White race and ethnicity was 313 (74.2%), and among the parents, 254 (60.2%) had a college degree or higher. Among the boys, the use of devices to calm at T2 was associated with higher emotional reactivity at T3 (r [standardized regression coefficient] = 0.20; 95% CI, 0.10-0.30), while higher emotional reactivity at T2 had a nonsignificant association with increased device use for calming at T3 (r = 0.10; 95% CI, -0.01 to 0.21). Among children with high temperamental surgency, the use of devices to calm at T2 was associated with increased emotional reactivity at T3 (r = 0.11; 95% CI, 0.01-0.22), while higher emotional reactivity at T2 was associated with increased device use for calming at T3 (r = 0.13; 95% CI, 0.02-0.24). **Conclusions and relevance:** The findings of this study suggest that the frequent use of mobile devices for calming young children may displace their opportunities for learning emotion-regulation strategies over time; therefore, pediatric health care professionals may wish to encourage alternate calming approaches.

Conflict of Interest Disclosures: Dr Radesky reported receiving personal fees from Noggin (Viacom/CBS) for serving on their scientific advisory board in 2021, and consulting fees from Melissa & Doug Toys outside the submitted work. Dr Kaciroti reported receiving grants from the University of Michigan during the conduct of the study. No other disclosures were reported.

Sensitivity and Specificity of the Modified Checklist for Autism in Toddlers (Original and Revised): A Systematic Review and Meta-analysis

Andrea Trubanova Wieckowski; Lashae N Williams; Juliette Rando; Kristen Lyall; Diana L Robins

JAMA Pediatr 2023 Apr 1;177(4):373-383.
doi:10.1001/jamapediatrics.2022.5975.

Abstract

Importance: The Modified Checklist for Autism in Toddlers (M-CHAT) and the M-CHAT, Revised With Follow-up (M-CHAT-R/F)-henceforth referred to as M-CHAT(-R/F)-are the most commonly used toddler screeners for autism spectrum disorder (ASD). Their use often differs from that in the original validation studies, resulting in a range of estimates of sensitivity and specificity. Also, given the variability in reports of the clinical utility of the M-CHAT(-R/F), researchers and practitioners lack guidance to inform autism screening protocols. **Objective:** To synthesize variability in sensitivity and specificity of M-CHAT(-R/F) across multiple factors, including procedures for identifying missed cases, likelihood level, screening age, and single compared with repeated screenings. **Data sources:** A literature search was conducted with PubMed, Web of Science, and Scopus to identify studies published between January 1, 2001, and August 31, 2022. **Study selection:** Articles were included if the studies used the M-CHAT(-R/F) (ie, original or revised version) to identify new ASD cases, were published in English-language peer-reviewed journals, included at least 10 ASD cases, reported procedures for false-negative case identification, screened children by 48 months, and included information (or had information provided by authors when contacted) needed to conduct the meta-analysis. **Data extraction and synthesis:** The systematic review and meta-analysis was conducted within the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) reporting guideline. The Quality Assessment of Diagnostic Accuracy Studies-2 tool evaluated bias in sample selection. Data extraction and quality assessment were performed by two authors independently. The overall diagnostic accuracy of the M-CHAT(-R/F) was assessed with the hierarchic summary receiver operating characteristic (HSROC) model. **Main outcomes and measures:** Sensitivity, specificity, diagnostic odds ratios, and HSROC curves of M-CHAT(-R/F). **Results:** The review included 50 studies with 51 samples. The pooled sensitivity of M-CHAT(-R/F) was 0.83 (95% CI, 0.77-0.88), and the pooled specificity was 0.94 (95% CI, 0.89-0.97). Heterogeneity analyses revealed greater diagnostic accuracy for low- vs high-likelihood samples, a concurrent vs prospective case confirmation strategy, a large vs small sample size, use of M-CHAT(-R/F) Follow-up, and non-English vs English only. **Conclusions and relevance:** Overall, results of this study suggest the utility of the M-CHAT(-R/F) as an ASD screener. The wide variability in psychometric properties of M-CHAT(-R/F) highlights differences in screener use that should be considered in research and practice.

Conflict of Interest Disclosures: Dr Wieckowski reported receiving grants from the Pennsylvania Medical Society and the Eagles Autism Foundation. Dr Lyall reported receiving grants from the Eagles Autism Foundation. Dr Robins reported receiving personal fees from M-CHAT LLC co-ownership, in which licensees pay royalties; receiving grants from the Eagles Autism Foundation, the National Institutes of Health, and the Pennsylvania Medical Society; having a contract to contribute to a Food and Drug Administration trial from Autism Speaks; receiving a gift to support pilot research from the Wawa Foundation; receiving personal fees from Quadrant Biosciences, Inc, for serving as a member of an advisory board; having a contract to collaborate on a toddler screening study in Monterrey, Mexico, from Autismo ABP outside the submitted work; and holding a copyright for M-CHAT, M-CHAT-R/F issued to M-CHAT, LLC (M-CHAT and M-CHAT-R/F are copyrighted instruments). No other disclosures were reported.

Outdoor Play as a Mitigating Factor in the Association Between Screen Time for Young Children and

JOURNAL ABSTRACTS

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

Neurodevelopmental Outcomes

Mika Sugiyama; Kenji J Tsuchiya; Yusuke Okubo; Mohammad Shafiur Rahman; Satoshi Uchiyama; Taeko Harada; Toshiki Iwabuchi; Akemi Okumura; Chikako Nakayasu; Yuko Amma; Haruka Suzuki; Nagahide Takahashi; Barbara Kinsella-Kammerer; Yoko Nomura; Hiroaki Itoh 8; Tomoko Nishimura

JAMA Pediatr., 2023 Mar 1;177(3):303-310.
doi: 10.1001/jamapediatrics.2022.5356.

Abstract

Importance: Whether the association between higher screen time in infancy and later suboptimal neurodevelopment can be mitigated by frequency of outdoor play is unknown. **Objective:** To investigate whether higher screen time at age two years is associated with neurodevelopmental outcomes at age four years and whether this association is mediated by frequency of outdoor play at age two years eight months. **Design, setting, and participants:** Participants were a subsample of the Hamamatsu Birth Cohort Study for Mothers and Children (HBC Study, N = 1258). Children were born between December 2007 and March 2012 and followed up from one year six months to four years. The analysis was conducted from April 2021 to June 2022. **Exposures:** Screen time longer than 1 hour a day at age two years was coded as higher screen time. **Main outcomes and measures:** Standardized scores for communication, daily living skills, and socialization domains of the Vineland Adaptive Behavior Scale, second edition, at age four years were used (mean [SD], 100 [15]). The mediating factor was frequency of outdoor play at age two years eight months, with six or sven days per week coded as frequent outdoor play. **Results:** Of 885 participants, 445 children (50%) were female; mean (SD) screen time per day was 2.6 (2.0) hours. Causal mediation analyses revealed that higher screen time at age 2 years was associated with lower scores in communication at age 4 years (nonstandardized coefficient b = -2.32; 95% CI, -4.03 to -0.60), but the association was not mediated by frequency of outdoor play. Higher screen time was also associated with lower scores in daily living skills (b = -1.76; 95% CI, -3.21 to -0.31); 18% of this association was mediated by frequency of outdoor play. Frequency of outdoor play was associated with socialization (b = 2.73; 95% CI, 1.06 to 4.39), whereas higher screen time was not (b = -1.34; 95% CI, -3.05 to 0.36). **Conclusions and relevance:** Higher screen time at age two years was directly associated with poorer communication at age four years. It was also associated with daily living skills, but frequency of outdoor play at age two years eight months alleviated it, suggesting outdoor play mitigated the association between higher screen time and suboptimal neurodevelopment. Future research should specify the nature of the associations and intervention measures, enabling targeted interventions that reduce the potential risk in screen time.

Physical Activity Interventions to Alleviate Depressive Symptoms in Children and Adolescents: A Systematic Review and Meta-analysis

Francesco Recchia; Joshua D K Bernal; Daniel Y Fong; Stephen H S Wong; Pak-Kwong Chung; Derwin K C Chan; Catherine M Capio; Clare C W Yu; Sam W S Wong; Cindy H P Sit; Ya-Jun Chen; Walter R Thompson; Parco M Siu

JAMA Pediatr. 2023 Feb 1;177(2):132-140.
doi: 10.1001/jamapediatrics.2022.5090.

Abstract

Importance: Depression is the second most prevalent mental disorder among children and adolescents, yet only a small proportion seek or receive disorder-specific treatment. Physical activity interventions hold promise as an alternative or adjunctive approach to clinical treatment for depression. **Objective:** To determine the association of physical activity interventions with depressive symptoms in children and adolescents. **Data sources:** PubMed, CINAHL, PsycINFO, EMBASE, and SPORTDiscus were searched from inception to February 2022 for relevant studies written in English, Chinese, or Italian. **Study selection:** Two independent researchers selected studies that assessed the effects of physical activity interventions on depressive symptoms in children and adolescents compared with a control condition. **Data extraction and synthesis:** A random-effects meta-analysis using Hedges g was performed. Heterogeneity, risk of bias, and publication bias were assessed independently by multiple reviewers. Meta-regressions and sensitivity analyses were conducted to substantiate the overall results. The study followed the PRISMA reporting guideline. **Main outcomes and measures:** The main outcome was depressive symptoms as measured by validated depression scales at postintervention and follow-up. **Results:** Twenty-one studies involving 2441 participants (1148 [47.0%] boys; 1293 [53.0%] girls; mean [SD] age, 14 [3] years) were included. Meta-analysis of the postintervention differences revealed that physical activity interventions were associated with a reduction in depressive symptoms compared with the control condition (g = -0.29; 95% CI, -0.47 to -0.10; P = .004). Analysis of the follow-up outcomes in 4 studies revealed no differences between the physical activity and control groups (g = -0.39; 95% CI, -1.01 to 0.24; P = .14). Moderate study heterogeneity was detected (Q = 53.92; df = 20; P < .001; I2 = 62.9% [95% CI, 40.7%-76.8%]). The primary moderator analysis accounting for total physical activity volume, study design, participant health status, and allocation and/or assessment concealment did not moderate the main treatment effect. Secondary analyses demonstrated that intervention (ie, <12 weeks in duration, 3 times per week, unsupervised) and

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

participant characteristics (ie, aged ≥13 years, with a mental illness and/or depression diagnosis) may influence the overall treatment effect. **Conclusions and relevance:** Physical activity interventions may be used to reduce depressive symptoms in children and adolescents. Greater reductions in depressive symptoms were derived from participants older than 13 years and with a mental illness and/or depression diagnosis. The association with physical activity parameters such as frequency, duration, and supervision of the sessions remains unclear and needs further investigation.

Association of Habitual Checking Behaviors on Social Media With Longitudinal Functional Brain Development
Maria T Maza; Kara A Fox; Seh-Joo Kwon; Jessica E Flannery; Kristen A Lindquist; Mitchell J Prinstein; Eva H Telzer
JAMA Pediatr. 2023;177(2):160-167.
doi:10.1001/jamapediatrics.2022.4924.

Key Points
Question Is adolescents' frequency of checking behaviors on three social media platforms (Facebook, Instagram, Snapchat) associated with longitudinal changes in functional brain development across adolescence. **Findings:** In this cohort study of 169 sixth- and seventh-grade students, participants who engaged in habitual checking behaviors showed a distinct neurodevelopmental trajectory within regions of the brain comprising the affective salience, motivational, and cognitive control networks in response to anticipating social rewards and punishments compared with those who engaged in nonhabitual checking behaviors. **Meaning:** These results suggest that habitual checking of social media in early adolescence may be longitudinally associated with changes in neural sensitivity to anticipation of social rewards and punishments, which could have implications for psychological adjustment.

Abstract
Importance: Social media platforms provide adolescents with unprecedented opportunities for social interactions during a critical developmental period when the brain is especially sensitive to social feedback. **Objective:** To explore how adolescents' frequency of checking behaviors on social media platforms is associated with longitudinal changes in functional brain development across adolescence. **Design, Setting, and Participants:** A 3-year longitudinal cohort study of functional magnetic resonance imaging (fMRI) among sixth- and seventh-grade students recruited from three public middle schools in rural North Carolina. **Exposures:** At wave 1, participants reported the frequency at which they checked Facebook, Instagram, and Snapchat. **Main Outcome or Measure:** Neural responses to the Social Incentive Delay task when anticipating receiving social feedback, measured annually using fMRI for three years. Participants saw a cue that indicated whether the social feedback (adolescent faces with emotional expressions) would be a reward, punishment, or neutral; after a delay, a target appeared and students responded by pressing a button as quickly as possible; a display of social feedback depended on trial type and reaction time. **Results:** Of 178 participants recruited at age 12 years, 169 participants (mean [SD] age, 12.89 [0.58] years; range, 11.93-14.52 years; 91 [53.8%] female; 38 [22.5%] Black, 60 [35.5%] Latinx, 50 [29.6%] White, 15 [8.9%] multiracial) met the inclusion criteria. Participants with habitual social media checking behaviors showed lower neural sensitivity to social anticipation at age 12 years compared with those with nonhabitual checking behaviors in the left amygdala, posterior insula (PI), and ventral striatum (VS; ß, -0.22; 95% CI, -0.33 to -0.11), right amygdala (ß, -0.19; 95% CI, -0.30 to -0.08), right anterior insula (AI; ß -0.23; 95% CI, -0.37 to -0.09), and left dorsolateral prefrontal cortex (DLPFC; ß, -0.29; 95% CI, -0.44 to -0.14). Among those with habitual checking behaviors, there were longitudinal increases in the left amygdala/PI/VS (ß, 0.11; 95% CI, 0.04 to 0.18), right amygdala (ß, 0.09; 95% CI, 0.02 to 0.16), right AI (ß, 0.15; 95% CI, 0.02 to 0.20), and left DLPFC (ß, 0.19; 95% CI, 0.05 to 0.25) during social anticipation, whereas among those with nonhabitual checking behaviors, longitudinal decreases were seen in the left amygdala/PI/VS (ß, -0.12; 95% CI, -0.19 to -0.06), right amygdala (ß, -0.10; 95% CI, -0.17 to -0.03), right AI (ß, -0.13; 95% CI, -0.22 to -0.04), and left DLPFC (ß, -0.10, 95% CI, -0.22 to -0.03). **Conclusions and Relevance:** The results of this cohort study suggest that social media checking behaviors in early adolescence may be associated with changes in the brain's sensitivity to social rewards and punishments. Further research examining long-term associations between social media use, adolescent neural development, and psychological adjustment is needed to understand the effects of a ubiquitous influence on development for today's adolescents.

Correction, February 13, 2023
Errors in Figures, *JAMA Pediatr*. 2023;177(4):440. doi:10.1001/jamapediatrics.2022.4924
In the article titled "Association of Habitual Checking Behaviors on Social Media With Longitudinal Functional Brain Development," published online January 3, 2023, in JAMA Pediatrics, the legends for the graphs in Figures 1 through 4 incorrectly labeled the participants' checking behavior. The green indicates moderate checking behavior, and the orange indicates low (nonhabitual) checking behavior. The article has been corrected.