Common Manual Therapy Practices in the Netherlands for Infants With Upper Cervical Dysfunction: A Prospective Cohort Study

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Abstract

Objective: The purpose of this study was to describe common clinical practices of manual therapists (MTs) in the Netherlands for infants with indications of upper cervical dysfunction (UCD).

Methods: A prospective observational cohort study was conducted to gain insight into characteristics, reasons for seeking care, and common clinical practice for infants (<27 weeks) with indications of UCD, referred to MTs. Pre- and posttreatment self-reported questionnaires were used to collect data from parents and MTs. Parents reported on infant characteristics and perceived effect of treatment. Manual therapists reported on diagnostics, therapeutic procedures, and outcomes.

Results: Between 2006 and 2007, data regarding 307 referred infants (mean age: 11.2 weeks) were collected by parents and 42 MTs. The most frequent reasons for seeking care were positional preference, restlessness, and/or abnormal head position. Manual therapists observed active, spontaneous, and provoked mobility and passive upper cervical mobility. Of the 307 infants, 295 were diagnosed with UCD based on positive outcomes on the flexion-rotation test and/or lateral flexion test. After treatment with mobilization techniques, positive outcomes on the flexion-rotation test decreased from 78.8% to 6.8%. For the lateral flexion test, the positive outcomes decreased from 91.5% to 6.2%. All parents perceived positive treatment effects. No serious adverse events were reported during this study.

Conclusions: This is the first study to describe common clinical practice for infants referred for manual therapy. Infants with UCD were treated mainly with upper cervical mobilization techniques, and the greatest perceived effect was observed after approximately 2 treatment sessions. (J Manipulative Physiol Ther 2018;41:52-61)

Key Indexing Terms: *Physical Therapy Modalities; Musculoskeletal Manipulation; Infant*

Introduction

Many infants are treated by manual therapists. In the Netherlands, manual therapists (MTs) are physical therapists with an additional postgraduate clinical education in manual therapy. Some MTs have received complementary education concerning manual therapy in infants and register their treatment sessions yearly.[1] Between 2006 and 2015, almost 75 000 infants were treated by MTs for posture asymmetry and/or dysfunction of the upper cervical spine.[2] Manual therapists apply only gentle spinal mobilization techniques; high-velocity thrust manipulation is avoided because of the risk of adverse events.[3,4] Although a large number of infants are referred for manual therapy yearly,[2] the therapeutic procedure and process of clinical reasoning in common practice are still scarcely described.

In the Netherlands, the therapeutic approach is based mainly on the KISS (kinematic imbalance due to suboccipital strain) concept, originally described by Gutmann as "diencephalon syndrome,"[5] and described as KISS by Biedermann.[6] The literature describes the KISS concept as a reversible biomechanical upper cervical dysfunction. This dysfunction is regarded as the underlying cause of reduced mobility in the upper cervical spine,[7] and would lead to an antalgic and biomechanical positional preference (PP) of the head and postural asymmetry.[6-10] In addition, the biomechanical upper cervical dysfunction results in reduced

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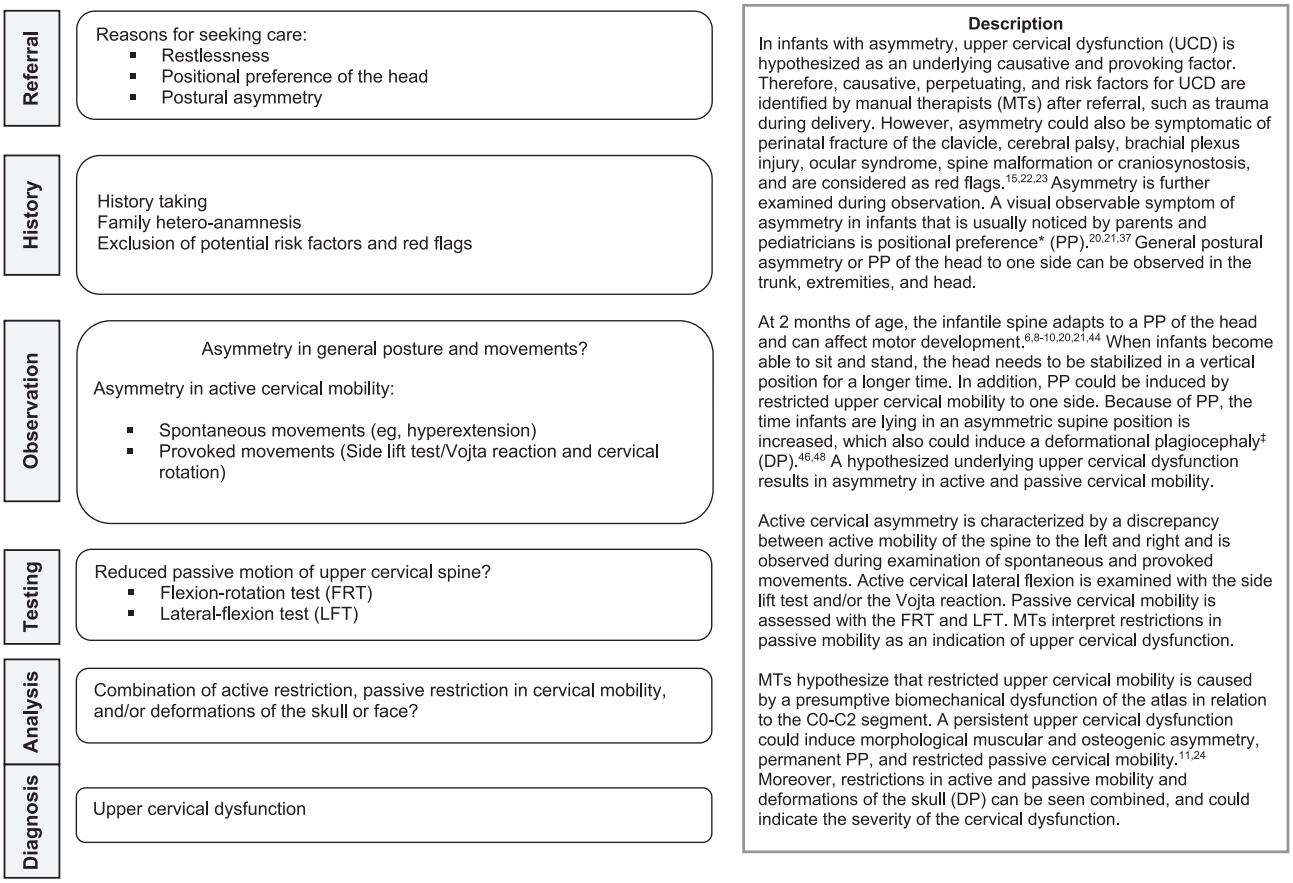

Fig 1. *Model of clinical reasoning of manual therapists in the treatment of infants. *PP (positional preference) is defined as showing head rotation to either the right or left side in supine position for a minimal three-quarters of a day, and which can be observed for 15 minutes during examination, combined with no active rotation of the head over the full 180º. ‡DP (deformational plagiocephaly) is defined as observed head and/or facial asymmetry and flattening of the occiput, as a result of pre- and/or postnatal forces on the growing skull, and cervical rotation restriction toward the nonflattened side.*

active and passive cervical joint mobility.[10] Upper cervical dysfunction (UCD) can be described as a fixed posture toward lateral flexion and contralateral rotation, combined with passive and active restricted cervical range of motion.[7] The underlying rationale in clinical reasoning is provided in Figure 1. However, evidence validating the assumptions is scarce. In this study, we describe UCD as a biomechanical dysfunction of the upper cervical spine and explore its consequences in infants.

The aim of this prospective cohort study was to gain insight into the patient characteristics and reasons for seeking care in infants with indications of UCD referred for manual therapy and to describe the diagnostic and therapeutic procedures and treatment outcomes in a transparent way. This study focused only on manual therapy in infants as a treatment for posture- and movement-related disorders from a primary biomechanical point of view. Therapeutic procedures based on different underlying constructs for treatment, such as chiropractic and osteopathy, fall outside the scope of this article. This article describes clinical practice in manual therapy in infants in the Netherlands and does not draw any conclusion about effectiveness.

Methods

Study Design and Procedure

This descriptive study was based on prospective observational data sampled from a clinical cohort between 2006 and 2007, collected by parents and MTs. All registered MTs (n = 239) specially educated in treatment of infants[1] were invited to participate in the study and asked to recruit parents/caregivers of referred infants. Participating MTs informed parents/caregivers ("parents") about the study, including the option to withdraw without consequences from treatment of the infant. Informed consent had to be signed by parents prior to study participation. The ethics review and approval for this study was completed by the Dutch Central Committee of Research Involving Human Subjects (CCMO: dd. March 15, 2007).

Participants

Included were infants aged <27 weeks referred with indications of UCD and without causative concomitant pathology, potential underlying pathology, and/or red flags in the referral information or history taking.[11] The infants received no other treatment at the symptom or dysfunctional level.[11] Only pediatric physical therapy was allowed because it is usual care in these infants and because of its focus on symptoms at the level of activities and participation; no treatment at the biomechanical level was performed.

Measurements

Three MTs (E.R.I.C.S., B.H.v.d.W., T.S.), 2 pediatric physical therapists (E.D., P.D.), a pediatric physician (G.S.), and an obstetrician (G.S.W.) developed 2 descriptive questionnaires to gain information on symptoms and risk factors for UCD based on existing literature[5,7,9] and clinical expertise. In addition, these self-reported questionnaires were used to assess diagnostics, treatment procedures, outcomes, and harms. Harms were classified as *mild* (transient side effect lasting <24 hours), *moderate* (requiring medical and/or general practitioner treatment), and *severe* (requiring hospital treatment, adverse event).[12] The concept questionnaires were substantively discussed by 10 MTs and consensus was reached on 2 questionnaires: 1 for parents and 1 for MTs. The MTs received all the questionnaires from the research team by post. Each questionnaire consisted of 2 sections: One collected baseline measurements; the other was used for posttreatment evaluation. Before treatment, MTs handed out the questionnaire with 2 stamped self-addressed envelopes to parents. Parents completed the pretreatment section and returned the questionnaire directly to the research team. After the final treatment session, parents completed the posttreatment section and returned it in the envelope. MTs completed their questionnaire during the first treatment session and after the last session, independently of the parents. Consequently, parents and MTs were blinded to each other's answers at both measurement moments and returned their questionnaires separately to the research team, to minimize risk of bias. The national register of MTs required that the occurrence of moderate or severe harms and adverse events be reported immediately.

All questionnaire sections were anonymized and were encoded with a unique number. Only an independent research assistant could match MTs' and parents' questionnaires by encoding.

Questionnaire for Parents. The pretreatment questionnaire given to parents included questions on infant characteristics, such as referral type, demographics, and reasons for seeking care, and information about the pregnancy and delivery. The second questionnaire was aimed at evaluating the infants' health status, treatment outcomes, and days between treatment and first perceived effect. A Global Perceived Effect (GPE) scale was added to assess the perceived treatment effect of parents on symptoms (1 = very much improved or totally cured; 2 = much improved; 3 = minimally improved; 4 = no change; 5 = minimally worse; 6 = much worse; 7 = very much worse or worse than ever),[13,14] and a visual analogue scale (VAS) was used to measure reduction in symptoms of the reason for seeking care over time, where scores of 0 and 100 indicate no and maximum change, respectively.

Questionnaire for MTs. In the first section, MTs were asked to report on the diagnostic process by describing observations of posture and movement, physical examination, and pretreatment measurements. The second section was used to report on treatment information, such as number of treatment sessions, applied techniques, posttreatment measurements, multidisciplinary collaboration, and harms. All questionnaires are available in Appendix A.

Clinical Outcomes. During clinical examination, MTs observe postural asymmetry and spontaneous and provoked movements to indicate possible underlying pathology.[15-17] To exclude anomalies, abnormalities, or underlying pathology, MTs use a standardized checklist of red flags.[11] When red flags are signaled, infants are referred back to the pediatrician or general practitioner. Importantly, trauma, presumed spine anomalies, congenital abnormalities, medullary tumors, neoplasms, unilateral visual dysfunction, or other concomitant pathologies can also induce cervical dysfunction and PP,[15,16,18-20] and have to be excluded. MTs perform neurologic screening to examine neuromusculoskeletal system functioning and development by testing reflexes and reactions.[17,21] Subsequently, active mobility of the upper cervical spine toward lateral flexion is examined using the side lift test and/or Vojta reaction.[17,21] To measure the passive mobility of the upper cervical spine, the flexion-rotation test (FRT) and lateral flexion test (LFT) are used. Detailed descriptions of these tests are provided in Appendix B. Positive scores on these tests indicate reduced cervical mobility and are interpreted as an indication of UCD. Both active mobility and passive mobility are tested, because they can occur independently of each other; reduced active mobility can be found without a reduction in passive mobility. To evaluate treatment outcome, assessment of cervical mobility using the FRT and LFT is repeated during the treatment process. To date, the reliability and validity of these upper cervical tests and mobility assessment have been studied for adults only.[22-24] In infants, only intrarater reliability has been indicated,[25] but validity and interrater reliability are still unknown.

Therapeutic Procedure (Treatment)

In the Netherlands, the manual therapeutic approach for infants differs slightly from the procedure described in other studies and countries.[26-28] To increase mobility and abrogate biomechanical dysfunction, only mild mobilization techniques are recommended, which focus primarily on

dysfunction of the atlas (C1) in relation to C0-C2, whereas high-velocity thrust manipulation is avoided. Previous literature has acknowledged the variation in applied amplitude between pediatric patients and adults[12,29] and described applied forces on infants with an average impulse of 30 to 70 N.[30] In contrast, Dutch MTs apply mobilization techniques with a very low level of force. In line with recent recommendations to use techniques with a maximum of 20 N in infants,[29] Dutch MTs are trained to apply manual pressure of about 11 N instead. Moreover, parents receive postural advice. Based on clinical reasoning and treatment outcomes, MTs decide how many treatment sessions are provided. Detailed information concerning upper cervical techniques is described in Appendix B.

Statistical Analysis

Descriptive statistics were used to describe infant characteristics and to evaluate diagnostic and treatment outcomes. χ^2 tests were used to compare these dichotomous outcomes between groups. Answers of "not applicable" were interpreted as missing values. Data were analyzed using SPSS, Version 20 (IBM, Armonk, New York).

Results

Of the 239 invited MTs, 78 (32.6%) were interested in participating in the study, and 42 (17.6%) collected data. Between September 2006 and September 2007, questionnaires on 331 infants were completed and returned (mean: 8 infants per MT, range: 1–36). Of these, 24 were older than 27 weeks at baseline and, therefore, were excluded from further analysis. The total study population consisted of 307 infants.

Characteristics and Reasons for Seeking Care

Table 1 lists the characteristics of these 307 infants. The mean age was 11.2 weeks (SD: 5.2, range: 2.0–26.0), and 62.3% were boys. One-third of the parents contacted MTs directly using the method of direct-access physical therapy, whereas the other infants were predominantly referred by pediatric physical therapists (31.1%) or a pediatrician or midwife (22.1%). The most frequently reported reasons for parents to seek care were clear positional preference (79.0%), restlessness (61.0%), and abnormal head position (54.8%). The most frequently reported disturbing symptoms were positional preference (28.7%), excessive crying (25.1%), and abnormal head position (18.8%).

Clinical Outcomes

During observation, MTs reported PP in 273 infants (88.9%). In these 273 infants, mainly a right rotation-left lateral flexion preference was observed (46.4%), followed by left rotation-right lateral flexion (28.5%). As outlined in Table 2, in 54.1% of the infants, a deformational plagiocephaly (DP) was observed, and hyperextension was observed in 67.1% of the infants. When infants were subdivided into infants with PP (n = 273) and infants without PP (n = 34), significant differences in observations were revealed. The prevalence of DP was statistically

Table 1. *Characteristics of the Study Population (n = 307)*

Characteristic	N (%)[a]
Mean age, wk (SD) (missing[b] n = 0)	11.2 (5.2)
Sex (missing n = 3)	
Boys	192 (62.5)
Girls	112 (36.5)
First-born infants (missing n = 1)	142 (46.3)
Second-born infants (missing n = 1)	125 (40.7)
Referrer (missing n = 18)	
General practitioner	21 (6.8)
Pediatrician/midwife	64 (20.8)
Pediatric physical therapist	92 (30.0)
Direct-access physical therapy	95 (30.9)
Other referrer	17 (5.5)
Symptoms, reported by parents (missing n = 2)	
Deviating position of the head	167 (54.4)
Deviating position of the trunk	88 (28.7)
Excessive crying	152 (49.5)
Fearfulness	164 (53.4)
Restlessness	186 (60.6)
Sleep disturbance	124 (40.4)
Obvious positional preference	241 (78.5)
Characteristics of pregnancy and delivery	
Infants from multiple pregnancy (missing n = 3)	14 (4.6)
Abnormal position in utero (missing n = 2)	60 (19.5)
Maternal complications during pregnancy (missing n = 1)	90 (29.3)
Mean pregnancy duration, wk (missing n = 0)	39
Deliveries with assistance of (missing n = 0)	
Midwife	156 (50.7)
Gynecologist	137 (44.5)
Other	14 (4.8)
Abnormal course of delivery (missing n = 2)	
Vacuum extraction or traction on the head	91 (29.6)
Cesarean	33 (10.7)
Forceps delivery	5 (1.6)
Postnatal complications in infants (missing n = 12)	
Meconium in amniotic fluid	33 (10.7)
Umbilical cord around the neck	60 (19.5)
Bone fractures after birth	5 (1.6)
Hematomas after birth	42 (13.7)
Mean second Apgar score >7 (SD) (missing n = 53)	1 (0.3)

SD, standard deviation.
[a] Unless noted otherwise.
[b] Values are defined as missing when parents did not complete the item on the questionnaire or answered with "don't know" or "inapplicable."

Table 2. *Outcomes of the Observation by Manual Therapists of the Total Study Population of Infants, Classified With and Without PP (Total Study Population, n = 307)*

	Total Observed (n)	Positive Outcome (n)	Percentage
Deformational plagiocephaly	303	164	54.1%
Hyperextension	301	202	67.1%
Facial asymmetry	306	60	19.6%
Fixed posture of the head	283	89	31.4%

	With Observed PP (n = 273)		Without Observed PP (n = 34)		
	Positive outcome (n)	Percentage	Positive outcome (n)	Percentage	*P* value [a]
Deformational plagiocephaly	159	58.2%	5	14.7%	<.001
Hyperextension	176	64.5%	26	76.5%	—
Facial asymmetry	60	22.0%	0	0%	.009
Fixed posture of the head	89	32.6%	0	0%	<.001

PP, positional preference.

[a] The *P* value was based on χ^2 tests between infants with observed PP and infants without observed PP. A difference was considered statistically significant at $P < .05$.

significantly lower in infants without PP ($P < .001$) than among infants with PP. The prevalence of hyperextension did not differ significantly between groups. With respect to facial asymmetry and a fixed posture of the head, infants without PP differed statistically significantly from infants with PP. In contrast to infants with PP, who did manifest facial asymmetry (22.0%) and a fixed posture (32.6%), these observations were absent in infants without PP.

Table 3 outlines the outcomes of tests performed by MTs before treatment and after the last treatment session. After treatment, the percentages of positive outcomes, indicating restriction in upper cervical mobility, decreased within all tests. Restrictions in active upper cervical lateral flexion, assessed with the side lift test and/or Vojta reaction, decreased by 51.1%. This decrease was congruent between infants with PP and those without PP.

The FRT and LFT were used to examine passive cervical mobility. Before treatment, 78.8% and 91.5% had positive outcomes on the FRT and LFT, respectively. After treatment, 6.8% had a positive outcome on the FRT, indicating a restriction in passive upper cervical mobility toward flexion and rotation; 6.2% had a positive LFT, indicating a restriction toward lateral flexion. These percentages were larger in infants with PP (n = 273): 80.6% had positive outcomes on the FRT, and 7.3% of the infants still exhibited this movement restriction after treatment. The LFT was positive in 92.7% of the infants and decreased to 6.6% after treatment. In infants without observed PP (n = 34), there were comparable decreases in positive outcomes on the FRT and LFT after treatment. Positive outcomes on the FRT decreased from 64.7% to 2.9%, whereas positive LFT outcomes decreased from 82.4% to 2.9%. Manual therapists reported a positive FRT and/or LFT in 295 of the 307 infants; these 295 infants were diagnosed with UCD. At the end of the treatment cycle, MTs reported that PP was absent in 98.2% of the infants.

Therapeutic Procedure

All 307 infants were treated by MTs. As outlined in Table 4, MTs mostly performed mobilizations toward lateral flexion of C1 (86.9%) and flexion of C0–C1 (66.1%). Infants diagnosed with UCD (n = 295) received treatment of the upper cervical spine and sacroiliac joint most frequently. Lateral mobilization of C1 was performed in 87.5% of the infants and in 88.6% of the infants with a positive LFT (n = 281). Other treatment techniques were flexion mobilization of C0–C1 (66.1%) and/or C1–C2 (37.6%), upper cervical traction (35.9%), and mobilization of the sacroiliac joint (53.6%). An additional mobilization toward flexion of C0–C1 and C1–C2 was performed in 68.2% and 39.3% of infants with a positive FRT (n = 242), respectively (data not shown). The average number of treatment sessions for infants treated for UCD was 3.5 (SD: 1.2, range: 1–6), with an average interval of 2.2 weeks (SD: 1.0) between sessions. Only a minority of infants (7.8%) received more than 5 treatment sessions. In the 12 infants without a diagnosis of UCD, mostly mobilizations of the upper cervical spine toward flexion and lateral flexion and of the sacroiliac joint were performed. Eight of these infants had an observable PP before treatment and were mobilized on the level of C0–C2 (n = 5) and C3–C7 (n = 1), total spine (n = 1); only 1 infant had anterior-posterior mobilizations of the spine. Violation of the proposed model of clinical reasoning was seen in 2 of 307 infants, in whom, despite negative tests and no observed PP, mobilizations of C0-C2 were still performed. The average number of treatment sessions for infants without UCD was 2.9 (SD: 0.64, range: 2–4), with an average treatment session interval of 2.9 weeks (SD: 1.5).

Harms

Vegetative responses after mobilization were reported: flushing (17.8%), hyperextension (4.3%), perspiration

Table 3. *Pre- and Post-treatment Outcomes in Infants, Reported by Manual Therapists (Total Study Population, n = 307)*

	Pretreatment		Post-treatment		
Performed test [a]	n	Percentage	n	Percentage	Delta
Side lift test and/or Vojta reaction (total)	231	75.2%	257	83.7%	+8.5%
Positive test	191	62.2%	34	11.1%	–51.1%
Flexion-rotation test (total)	299	97.4%	301	98.0%	+0.6%
Positive test	242	78.8%	21	6.8%	–70.0%
Lateral flexion test (total)	302	98.4%	301	98.0%	–0.4%
Positive test	281	91.5%	19	6.2%	–85.3%
Flexion-rotation test and/or lateral flexion test (total)	307	100%	307	100%	0%
Positive test	295	96.1%	25	8.1%	–88.0%

	Infants With Observed PP (n = 273)					Infants Without Observed PP (n = 34)				
	Pretreatment		Posttreatment			Pretreatment		Posttreatment		
Performed test [a]	n	Percentage	n	Percentage	Delta	n	Percentage	n	Percentage	Delta
Side lift test and/or Vojta reaction (total)	210	76.9%	233	85.3%	+8.4%	21	61.7%	24	70.6%	+8.9%
Positive test	171	62.6%	31	11.4%	–51.2%	20	58.6%	3	8.8%	–49.8%
Flexion-rotation test (total)	267	97.8%	267	97.8	0%	32	94.1%	34	100%	+5.9%
Positive test	220	80.6%	20	7.3%	–73.3%	22	64.7%	1	2.9%	–61.8%
Lateral flexion test (total)	271	99.3%	268	98.2%	–1.1%	31	91.2%	33	97.1%	+6.1%
Positive test	253	92.7%	18	6.6%	–86.1%	28	82.4%	1	2.9%	–79.5%
Flexion-rotation test and/or lateral flexion test (total)	273	100%	273	100%	0%	34	100%	34	100%	0%
Positive test	265 [b]	97.1%	23	8.4%	–88.7%	30 [b]	88.2%	2	5.9%	–82.3%

PP, positional preference.
[a] Manual therapists reported if they had performed the test and if the test was positive or not.
[b] Statistically significant difference between infants with observed PP and infants without observed PP ($P = .02$).

(3.6%), and gastroesophageal reflux (0.3%). These symptoms disappeared immediately. Short breathing pattern changes were reported in 9.2% after mobilization and were classified as mild, transient side effects [mean duration: 2.5 seconds; range: 1–5]. No moderate or severe harms or serious adverse events were reported.

Therapeutic Collaboration

For 69.1% of the 307 infants, MTs reported therapeutic collaboration with (allied) health professionals. In the treatment of 147 infants (47.9%), MTs collaborated with a pediatric physical therapist. Other health professionals who collaborated were pediatricians (19.2%) and general practitioners (15.0%).

Perceived Effect

After treatment, parents reported much (51.7%) and large (40.7%) improvement of symptoms on the GPE; 7.6% reported minimal improvement. Perceived reduction in symptoms, as measured with the VAS, was 87.5 (SD: 11.8; range: 28-100). Among infants with pretreatment observed PP (n = 273), 35% of the parents still reported the presence of PP after treatment; perceived reduction in symptoms was 82.7 (SD: 12.9; range: 34–100). On the GPE, these parents reported minimal (12.6%), much (66.3%), and large improvement (21.1%). All parents of the 8 infants without positive diagnostic tests and the 2 infants without positive tests and no PP reported reduced symptoms. Within 3 days after a treatment session, 74.0% of the parents (n = 307) perceived reduced symptoms, with positive treatment effects 1 day after treatment (32.9%) being noted most frequently. The largest treatment effect was observed after 1.8 (SD: 1.1) treatment sessions.

Discussion

This is the first study to describe common practice of manual therapy for infants referred with indications of UCD. Manual therapists observed active, spontaneous and provoked movement and passive upper cervical mobility, specifically. The 295 infants diagnosed with UCD were treated with upper cervical mobilization techniques, where lateral mobilization of the atlas was performed most frequently. After treatment, all parents perceived a reduction in symptoms, with the largest effect after 1.8 treatment sessions. In all infants, passive upper cervical mobility was increased after treatment. No adverse events have been reported.

The prevalence of PP in the included study population, especially among boys, was high. This finding is comparable to those of other studies in terms of the larger number of boys with PP.[31-33] Compared with the total Dutch population in the same

Table 4. *Therapeutic Techniques Performed by Manual Therapists in Infants (n = 307)*

Treatment Technique	n	Percentage
Traction C0–C1	107	34.9%
Flexion C0–C1	203	66.1%
Lateral mobilization C1	266	86.9%
C1–C2 mobilization	113	36.8%
Extension/lateral flexion C0–C3	97	31.6%
Anterior-posterior mobilizations spine	89	29.0%
Sacroiliac joint mobilization	165	53.7%
Hip joint mobilization	21	6.8%

	Infants Diagnosed With UCD (n = 295)		Infants Without Diagnosis of UCD (n = 12)		
Treatment technique	n	Percentage	n	Percentage	*P* value [a]
Traction C0–C1	106	35.9%	1	8.3%	.036
Flexion C0–C1	195	66.1%	8	66.7%	—
Lateral mobilization C1	258	87.5%	8	66.7%	.006
C1–C2 mobilization	111	37.6%	2	16.7%	—
Extension/lateral flexion C0–C3	95	32.2%	2	16.7%	—
Anterior-posterior mobilizations of spine	35	11.9%	3	25.0%	—
Sacroiliac joint mobilization	158	53.6%	7	58.3%	—
Hip joint mobilization	20	6.8%	1	8.3%	—

UCD, upper cervical dysfunction.

[a] The *P* value was based on χ^2 tests between infants with UCD diagnosis and infants without UCD diagnosis. A difference was considered statistically significant if $P < .05$.

year of the cohort, infants in our study were observed with an abnormal position *in utero* (19.6%) and complications during delivery (29.3%) more frequently, with prevalences in Dutch infants being 7.0% and 8.3%,[34] respectively. In addition, despite the increasing trend in the prevalence of cesarean deliveries in the total Dutch population (15.0%), fewer cesarean deliveries were reported in our study (10.7%), and 29.6% of all the infants in our study were delivered by vacuum extraction and traction deliveries, compared with 9.7% in the total population.[34]

Based on previous research indicating a relation of asymmetry and PP with malposition *in utero*, delayed delivery, and head trauma during delivery or shortly after birth,[6,35] our findings support the hypothesis that abnormal deliveries increase the risk of UCD and PP. However, the literature is inconclusive on this point, conflicting results are described,[18,36] and extensive and high-quality research on infants with UCD is currently lacking. Moreover, an abnormal delivery can also result in greater parental concerns and earlier admission to health care. In addition, infants with PP in our study were more frequently observed with facial asymmetry and DP than infants without PP. Research has indicated these symptoms are a direct effect of underlying upper cervical dysfunction,[6,7,9,37] and causality between PP and the development of DP is assumed.[31,32,35,38-40] In line with previous research,[41] our study indicated that PP was frequently combined with DP, and in infants without PP, the prevalence of DP was statistically significantly lower.

In our study, infants were relatively young (mean age: 11.2 weeks). Furthermore, in line with previous research, we found that a right rotational preference of the head was observed more than a preference for the left.[7,31,37] Research on the natural course of PP indicated that infants up to 12 weeks of age have a physiological rotational preference that can only be passively mobilized normally, whereas contralateral rotation can be performed actively over the total range.[35,42] In addition, the prevalence of PP in healthy infants increases to 16 weeks of age,[31] but could also disappear spontaneously without further treatment within months to years.[43] Another study compared the effect of pediatric physical therapy with usual care for DP in infants with PP, and found that after 4 months of pediatric physical therapy, the prevalence of DP was significantly decreased compared with that for usual care. Notably, 4 and 10 months after study onset, PP was absent in both treatment groups.[44] Therefore, it cannot be totally excluded that the observed PP is part of the normal development of infants and naturally decreases in time. However, infants in our study were frequently referred by health professionals who are educated in recognizing the difference between a physiological PP and PP caused by an assumed cervical dysfunction.

Interestingly, in our study, symptoms of PP were reduced after a minimum amount of treatment sessions: The greatest effects were perceived after 1.8 sessions and within 3 days after treatment. Positive treatment outcomes were perceived by parents and observed by MTs, independently. Parents reported improvement using the GPE and VAS, and in both groups, MTs reported improvement in cervical mobility. Remarkably, parents of infants with PP reported that infants still exhibited PP in 35% of cases, but GPE and VAS scores were high. The severity of PP was probably decreased, but other symptoms

could also be reduced by treatment. For example, infants could be referred for restlessness or abnormal head position, the other most frequently reported reasons for seeking care, which were probably reduced based on the high VAS scores reported by parents.

Prior to this study, we did expect that infants with PP would largely exhibit more movement restriction than infants without PP. In contrast, we found that both infants with and those without PP had restrictions in cervical mobility, and mobility was improved in both groups after treatment. To further investigate and explain this outcome, the reliability of the FRT and LFT should be assessed.

In the majority of infants (n = 295) referred for manual therapy, the restricted range of upper cervical motion was interpreted as supportive evidence for the KISS concept,[5-7] although published peer-reviewed literature is scarce and inconsistent about this concept.[35] A rotational preference and/or restriction is usually noticeable in the observation of infants with PP, whereas the KISS concept focuses primarily on a passive lateral flexion dysfunction at C0–C1.[7] However, our study indicates that in infants with reported symptoms of excessive crying and restlessness, but without observable PP, DP, and hyperextension, restricted upper cervical mobility was also frequently observed. The assumption that UCD is often combined with PP could therefore underestimate the total number of infants with UCD, in which symptoms of DP and hyperextension should also be taken into account. However, the high coincidence of PP and cervical mobility restrictions makes it difficult to distinguish between the 2 symptoms, which is reinforced by the fact that the diagnostic tests currently used in common clinical practice are not validated in infants yet. Contamination between observations and test outcomes can therefore not be excluded.

We observed that dysfunction in cervical flexion-rotation and lateral flexion did not always result in mobilization at the same spinal level. Although many infants had cervical rotational restrictions, MTs mostly performed lateral flexion and flexion mobilizations. This finding is consistent with the hypothesis that an upper cervical lateral flexion deviation is the underlying provoking factor in dysfunction of the upper cervical spine (Fig 1),[7,8] which explains the large number of lateral mobilizations performed among MTs. Remarkably, both the cervical lateral flexion and flexion-rotational mobility were increased after treatment. This improvement in rotational mobility, despite not being treated specifically, confirms the hypothesis of the strong upper cervical biomechanical link, indicating that segmental mobilization toward lateral flexion induces a rotational mobilization at the same time.[45] This is generally enough to block the multisegmental dysfunction and increase mobility without further treatment. We thereby postulate that rotational mobilization is usually avoided during treatment because of the higher risk of adverse events[3] and explain the hesitation to perform rotational mobilizations.

No serious adverse events were reported by MTs. In 9.2% of the infants, MTs reported short breathing changes, with a mean duration of 2.5 seconds. Because these changes lasted less than 10 seconds, they could not be classified as apnea, and were interpreted as a side effect.[46]

Strengths and Limitations

We are the first to describe common clinical practice in manual therapy in infants. To avoid reporting bias, MTs were blinded to parents' reports and vice versa. This study provides insight into characteristics of referred infants, diagnostic and therapeutic procedures, and the effects perceived by MTs and parents. These factors should be guiding principles in future more robust studies. Our study is only a starting point to gain insight into daily practice and the clinical reasoning followed by MTs and the effects perceived by parents.

In 69.1% of cases, MTs reported collaboration with other health professionals, specifically in relation to pediatric physical therapy, which normally consists of parental advice and treatment focused on the level of activity and participation instead of biomechanical functioning.[47] However, the rationale and/or content of this collaboration were not reported. Therefore, questions arise as to the extent to which manual therapy contributes to treatment effects. Outcomes as a result of natural course, co-intervention, Hawthorne, or placebo effects in parents and/or MTs cannot be excluded. The rapid perceived treatment effect could therefore not be explained. However, the aim of our study was to describe clinical practice in manual therapy in infants and not to investigate effectiveness.

A total of 239 specialized MTs were invited to participate in the study, but only 42 participated in the process of data collection. This makes it difficult to indicate the generalizability of our study population. Because we did not know how many parents were invited by MTs to participate and there was no dropout after the start of treatment in this study, we cannot exclude selection bias of included infants.

Although UCD is described as a dysfunction with restrictions in both active and passive mobility, our questionnaire for MTs lacked a question on active cervical rotational mobility. So we were not able to identify the prevalence of active rotational restrictions in infants with UCD.

To date, the clinically used FRT and LFT have not been tested for validity and reliability in the present study population. Future research is necessary to validate how these diagnostic measures affect treatment decisions and to examine interrater reliability.

Although many infants were clinically treated and positive treatment effects were observed, this observational study did not include a randomized design and control group. Thus, no conclusion can be drawn regarding the effectiveness of upper cervical manual therapy. Given its prevalent application, additional randomized controlled studies that provide

fundamental knowledge about manual therapy in infants with UCD and its effectiveness are necessary.

Conclusions

In the majority of infants referred for manual therapy, a PP and restricted passive upper cervical movement were found. In common clinical practice, active, spontaneous, and provoked cervical mobility of infants was observed. The majority of infants were treated with upper cervical mobilization techniques, with the greatest effect being perceived after approximately 2 treatment sessions. All parents reported reduction in symptoms, and upper cervical mobility assessed by MTs was substantially increased. No serious adverse events were reported.

Practical Applications

- In the majority of infants (mean age: 11.2 weeks) with upper cervical dysfunction referred for manual therapy, positional preference of the head and reduced active and passive mobility of the upper cervical spine were observed.
- After gentle upper cervical mobilization techniques, active and passive cervical mobility increased and parents reported a reduction in symptoms.
- No adverse treatment effects were reported.

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Contributorship Information

Concept development (provided idea for research): E.R.I.C.S., B.H.v.d.W.
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Supervision (provided oversight, responsible for organization and implementation): E.R.I.C.S., B.W., R.A.d.B., M.W.G.N.v.d.S.
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Analysis/interpretation (responsible for statistical analysis, evaluation, and presentation of the results): F.D.
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Writing (responsible for writing a substantial part of the manuscript): F.D., E.R.I.C.S.
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Supplementary data

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