

Manipulation techniques for infant torticollis

ORIGINAL ARTICLE

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BACKGROUND

Professionals in fields including chiropractic, physiotherapy, manual therapy and osteopathy recommend manipulation techniques for the treatment of infant torticollis. This article summarises the research evaluating such treatment.

MATERIAL AND METHOD

The article is based on a search in MEDLINE, EMBASE, CINAHL, Cochrane CENTRAL, AMED, PEDro, and the Index to Chiropractic Literature in August 2017. Controlled studies were included to assess efficacy, and observational studies to reveal the risk of harm.

RESULTS

We reviewed 3 418 unique hits from the literature search. We included three randomised controlled studies on the efficacy of manual therapy or osteopathy and three observational studies on possible adverse effects. Two studies on manual therapy suggested that manipulation techniques do not have any greater effect on symmetry and motor outcomes than parental guidance, physiotherapy and home exercises. One study suggested that osteopathy may contribute to increased symmetry compared to placebo, but the clinical significance of this change is uncertain. Neither the three randomised studies nor a large patient series involving 695 infants found manipulation techniques to be associated with a risk of serious adverse events, but three patient histories demonstrated that manipulation techniques may cause harm if the observed asymmetry has serious underlying causes.

INTERPRETATION

Manipulation techniques have not been shown to be efficacious in the treatment of infant torticollis, but the available evidence must be considered uncertain.

Main points

Manipulation techniques are used to counteract infant torticollis, but the use of such techniques is controversial

We performed a comprehensive literature review to identify and summarise research on the efficacy and safety of manipulation techniques

Research did not support the use of manipulation techniques in the treatment of infantile asymmetry, but there was uncertainty associated with the research findings

Torticollis is the collective term for asymmetries of the head and neck region. The prevalence of congenital muscular torticollis, which is often accompanied by asymmetric head shape, is estimated at about 1 % (1). A greater number of infants, around 10 % of all those under six months, have an asymmetric head shape that may be related to restricted range of motion of the neck and to a positional preference when supine (2). Figures from Statistics Norway and the Norwegian

Health Economics Administration show that approximately 8 % of all Norwegian infants are taken to manual therapists in their first year, often due to head and neck-related asymmetries (3).

Conservative treatment of congenital muscular torticollis and other forms of asymmetry in infants, hereafter referred to as infantile asymmetry, consists of parental guidance, physiotherapy and home exercises (4, 5, 6). The aim is to establish symmetrical movement patterns through massage, passive stretching exercises and stimulation interventions (4, 7).

In the 1990s, H. Biedermann introduced the term cervical joint-induced symmetry disorder (also known as KISS, *Kopfgelenk-induzierte Symmetrie-Störung*), and described a possible association between imbalances of the cervical vertebrae in infants, symmetry disorders and symptoms such as colic and sleep and eating disorders (8, 9, 10). Biedermann believed that such imbalances could be treated with manipulation, i.e., brief gentle pressure applied with the index finger to the cervical joints (8). In Norway, this form of treatment is offered by practitioners in chiropractic, physiotherapy, manual therapy and osteopathy, but the efficacy and safety of manipulation techniques, especially in infants, is disputed (11, 12).

In this article, we searched for controlled studies that compared manipulation techniques with other treatments offered to infants with torticollis. We focused in particular on whether manipulation techniques contribute to more symmetrical posture and movement patterns, but we also considered the impact of treatment on autonomic outcomes like sleep, crying and restlessness. When searching for evidence of possible adverse effects, we considered all types of studies, irrespective of design.

Material and method

This article is an update of a systematic review published by the Norwegian Knowledge Centre for the Health Services in 2009 (13). A project plan was published in August 2015, describing the intended article in detail (14). In addition to using the search results from the earlier review (13), we also performed a search covering the period from the original search to August 2017. We conducted searches in MEDLINE, EMBASE, CINAHL, Cochrane CENTRAL, AMED, PEDro and the Index to Chiropractic Literature. The search was conducted by combining text and keywords describing the population (e.g. 'KISS', 'torticollis', 'asymmetric neck') and intervention (e.g. 'manual therapy', 'manipulation', 'osteopathy') (see appendix on tidsskriftet.no). We also searched for studies in the reference lists of other relevant reviews and studies.

To investigate efficacy, we included only controlled studies that fulfilled the selection criteria in Table 1. However, it can be difficult to detect rare adverse effects and harms in controlled studies. To investigate safety, we therefore allowed the inclusion of other types of studies too (Table 1).

Table 1

Criteria used to select studies to investigate the efficacy and safety of treatment. Studies that did not fulfil all criteria with respect to either efficacy or safety were excluded.

	Efficacy	Adverse effects
Design	Randomised, quasi-randomised and non-randomised controlled studies	All, including case reports
Population	Infants (0–2 years) in whom cervical joint-induced symmetry disorder was suspected ¹	Infants (0–2 years) treated for cervical joint-induced symmetry disorder ¹
Intervention	Manual therapy, chiropractic, osteopathy and physiotherapy targeting the cervical spine	Manual therapy, chiropractic, osteopathy and physiotherapy targeting the cervical spine
Comparison	No treatment, or placebo or other forms of treatment not targeting the cervical joints	
Outcome	Degree of symmetry, sleep disorders, duration of crying, passive rotation, etc.	Respiratory problems, nerve damage, paralysis, death, etc.

¹Studies on manipulation therapy for infants with colic were excluded.

Two of the authors selected articles independently of one another. Titles and abstracts were first reviewed for all hits from the literature search. Full-text articles were retrieved if at least one author considered the abstract to be relevant with respect to the selection criteria. An article was included if both authors agreed that it was relevant. Disagreements were resolved through discussion.

For studies that compared groups, we evaluated the risk of bias related to patient selection, study conduct, detection, follow-up and reporting using the Cochrane Collaboration's tool (15). This entails evaluation of factors such as randomisation procedures, masking, and dropout analysis. Studies without group-wise comparisons, which were included to reveal possible harms, were considered to have a high risk of bias. We contacted the Norwegian System of Patient Injury Compensation to obtain statistics on injuries reported after treatment of the neck by a chiropractor, manual therapist or physiotherapist.

Dichotomous outcomes were presented as relative risk (RR), and continuous outcomes as mean differences (MD) with 95 % confidence intervals. We used GRADE (Grading of Recommendations Assessment, Development and Evaluation) to assess whether we had high, medium, low or very low confidence that the reported outcomes resembled the true outcomes (16).

Results

The literature search resulted in the inclusion of six publications (Figure 1). Three of these were randomised controlled trials, two of which examined the efficacy of manual therapy (17, 18) and one, osteopathy (19). These studies are described in Table 2, which also summarises our rating of the risk of bias. In brief, the studies of Haugen *et al.* (17) and Philippi *et al.* (19) were exposed to few systematic sources of error. The study of Cabrera-Martos *et al.* (18) was considered to have methodological limitations that may have affected its conclusions. The limitations were primarily related to the lack of parental blinding, and to the fact that children in the manual therapy group received more frequent follow-up than those in the

control group. These differences may have contributed to greater treatment compliance in the manual therapy group. Details of the therapeutic techniques are presented in Table 3.

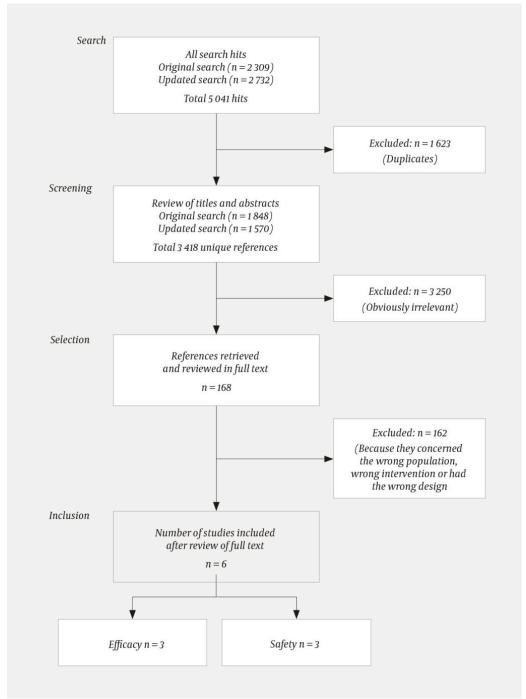


Figure 1 Flowchart summarising the selection process

Table 2

Characteristics of studies used to examine the efficacy and safety of manipulation techniques for infantile asymmetry. The studies are arranged within each category according to year of publication.

Study	Participants	Intervention	Control	Outcome	Risk GRADE of assessment ² bias ¹
Efficacy					

Study	Participants	Intervention	Control	Outcome	Risk of bias ¹	GRADE assessment ²
Cabrera- Martos et al. 2016 (18), randomised controlled trial	46 children with plagiocephaly, aged -5.7 months	Standard + manual therapy, weekly 45- minute sessions	Standard: Orthotic helmet + parental advice	Motor development: Satisfactory for all children Mean treatment duration: 110 and 149 days in intervention and control groups respectively, i.e. 39-day difference (95 % CI 31– 46)	High	For both outcome measures: ⊕⊙⊙⊙ Very low ^{3,4}
Haugen et al. 2011 (17), randomised controlled trial		Physiotherapy + manual therapy, two sessions of manual therapy	Physiotherapy alone	Change in symptoms ⁵ after 8 weeks: markedly fewer torticollis symptoms in 12/15 and 13/16 in intervention and control groups respectively. Relative risk 0.92 (95 % CI 0.16–5.49)	Low	⊕⊕⊙⊙ Low ³
Philippi et al. 2006 (19), randomised controlled trial	32 children with postural asymmetry, aged 6-12 weeks	Osteopathy, weekly treatment × 4	Placebo	Asymmetry ⁶ : Change in score was 4.7 points greater in intervention group than in control group (95 % CI 2.0-7.6).	Low	⊕⊕⊙⊙ Low ³
Safety						
Happle et al. 2009 (24), case report	2 children with postural asymmetry, aged 6 and 15 months	Manual therapy + physiotherapy	_	Delayed diagnosis Incorrect treatment	High	-
Koch et al. 2002 (23), patient series	695 children with postural asymmetry, aged 1– 12 months	Chiropractic	-	Heart rate	High	-

Study	Participants	Intervention	Control	Outcome	Risk GRADE of assessment ² bias ¹
Shafrir & Kaufman 1992 (25), case report	1 child with postural asymmetry, aged 4 months	Chiropractic	_	Delayed diagnosis Incorrect treatment Injury	High —

¹Randomised controlled studies were assessed using the Cochrane Collaboration's tool (15), while a discretionary judgement was made that patient series and case reports were at high risk of bias

Table 3Description of interventions used in the studies in this review.

Study	Description of intervention		
	Intervention group	Both groups	
Haugen et αl. 2011 (17)	Two treatments by manual therapist. During treatment, the head was held in a central position without stretching any structures. The atlas was gently manipulated in a direction appropriate to the functional impairment of the joint.	Advice and follow-up from a paediatric physiotherapist with the aim of stimulating equal movement on both sides. The physiotherapists had broad scope in deciding what content to include in the follow-up, but they were not to stretch the neck against resistance from the child.	
Cabrera-Martos <i>et al</i> . 2016 (18)	Weekly follow-up by manual therapist. The treatment was personalised, aimed to improve range of motion and function of the cervical spine, and predominantly targeted the spheno-occipital joint, atlanto-occipital joint and sacrum.	Advice on positioning and wearing an orthotic helmet 23 hours a day, plus monthly follow-up.	

²Grading of Recommendations Assessment, Development and Evaluation (16)

 $^{^3}$ Precision (-2), estimated efficacy has a very wide 95 % CI owing to the small number of participants

⁴Risk of bias (-1) because of possible age difference between groups, lack of parental masking, and different frequency of follow-up

⁵Physiotherapist's assessment of torticollis symptoms based on video recordings: worse, no change, better or much better

⁶Scale – developed by the study authors – from 4 (full symmetry) to 24 (maximum asymmetry) points based on evaluation of video recordings

Study	Description of intervention			
	Intervention group	Both groups		
Philippi et al. 2006 (19)	Four treatments by an osteopath who was free to use any techniques deemed necessary to correct the disorder. Osteopathy may consist of massage, manipulation techniques and stretching. The first author has established, in personal communication, that the techniques used bear many similarities to manual therapy.	Advice on positioning. Children in the control group underwent the same tests as those in the intervention group, but without receiving active treatment.		
Koch et al. 2002 (23)	One treatment by a chiropractor. During treatment, the infant was supine and all rotation of the spine was avoided. The therapist directed impulses (30 to 70 N) towards the suboccipital region on the side of the asymmetry.	_		
Happle et al. 2009 (24)	Manual therapy and physiotherapy (details not specified).	_		
Shafrir & Kaufman 1992 (25)	Chiropractic (details not specified).	_		

Manual therapy

Haugen *et al.* compared the efficacy of manual therapy as an adjunct to physiotherapy in infants aged between three and six months with reduced range of motion in the neck (17). The parents, the physiotherapists who performed the physiotherapy intervention and the individuals who evaluated the outcomes were unaware of which children had received manual therapy.

The children in the study were filmed at each visit. The recordings were used to quantify spontaneous motor function and strength as well as active and passive range of motion. The paediatric physiotherapist evaluated the final outcome by comparing recordings from the start of the study with those from the follow-up, and qualitatively assessing whether the child was worse, showed no significant change, was better or much better. After eight weeks, 12 of 15 participants in the treatment group and 13 of 16 in the control group were markedly better with fewer torticollis symptoms (RR 0.92, 95 % CI 0.16 to 5.49; see Table 2). There were no demonstrable differences between the groups in either symptom burden or in outcomes related to asymmetries in sleeping position, movement patterns, flexion or head rotation (17). Our confidence in the evidence is low for all outcomes owing to the small number of study participants and the imprecise effect estimates.

Cabrera-Martos *et al.* investigated the efficacy of manual therapy as an adjunct to parental guidance and treatment with an orthotic helmet <u>(18)</u>. The children's motor development was assessed using the Alberta Infant Motor Scale <u>(20)</u>, and asymmetry was classified according to the Argenta scale <u>(21)</u>. At the end of the study, asymmetry was minimal (0 or 1) and motor development satisfactory for all 46 children who participated in the study (Table 2). The mean treatment duration

in the control group was 149 days, which was 39 days longer (95 % CI 31–46) than for the children who received manual therapy. However, there is a high risk that the absence of parental blinding, and the fact that the children in the manual therapy group received more frequent follow-up than those in the control group, may have contributed to systematic biases.

Osteopathy

Philippi *et al.* randomised 32 infants with postural asymmetry to osteopathy or placebo (19). Children in the placebo group underwent the same tests as those in the intervention group but, for those in the placebo group, the osteopath placed his/her hands in the treatment position without performing active treatment.

The degree of symmetry was rated from 4 (full symmetry) to 24 (maximum asymmetry) using standardised video-based measurements (22). For participants in the control group, the symmetry score changed from 14.2 to 13.0, whereas for those in the osteopathy group, it changed from 15.4 to 9.5 (19). Children in the intervention group thus experienced an average improvement that was 4.7 points greater than in the control group (95 % CI 2.0 to 7.6 points; see Table 2). The authors did not provide effect estimates for the outcomes of vomiting, sleep, drinking, mood, excitability, defecation pattern and crying, but they found no statistically significant differences between the osteopathy and control groups (19). Due to the small number of study participants and imprecise effect estimates, we have low confidence in the evidence for all outcomes.

Risk of harm

The randomised studies included in this review did not report adverse effects, but we identified and included three descriptive studies – one patient series (23) and two case reports (24, 25) – to examine adverse effects and the risk of harm (Table 2).

Koch *et al.* presented data from 695 German infants who received manipulation treatment for cervical joint-induced symmetry disorders (23). No serious adverse effects were reported, but measurement of heart and respiratory rate revealed that manipulation treatment was often accompanied by transient apnoea and reduced heart rate, especially in the youngest children (23).

Happle *et al.* described two infants who were diagnosed with cervical joint-induced symmetry disorders and who were treated with manual therapy and physical therapy (24). Further investigation revealed that their symptoms, which persisted after completion of treatment, were due to cancer. The authors emphasised that infantile asymmetries may have serious differential diagnoses, and that delayed diagnosis can have serious consequences (24).

Shafrir & Kaufman described a four-month-old boy who was treated for infantile asymmetry by a chiropractor (25). After the manipulation, the boy quickly deteriorated. Examination revealed that the manipulation had triggered acute exacerbation of a previously undiagnosed intraspinal tumour (25).

Figures obtained from the Norwegian System of Patient Injury Compensation show that, since 2009, there have been no reports of injuries to Norwegian infants as a result of neck manipulations performed by chiropractors, manual therapists or physiotherapists. During the same period, 55 adult patients aged between 28 and

60 years sought compensation following neck manipulation by a chiropractor, manual therapist or physiotherapist. Eight had their claims upheld, including one in which the treatment was judged likely to have triggered a stroke.

Discussion

After reviewing the literature, we included three randomised controlled studies on the efficacy of manual therapy or osteopathy for infantile asymmetries of the head and neck. Haugen *et al.* showed that physiotherapy and manual therapy produced a change in symptoms similar to that seen with physiotherapy alone (17). Cabrera-Martos *et al.* suggested that manual therapy targeting the cervical spine may shorten treatment duration in infants with head asymmetries undergoing treatment with an orthotic helmet, but methodological limitations raise doubts about the result (18). Philippi *et al.* showed that a complex osteopathic intervention may potentially counteract asymmetry in infants with postural asymmetry, but found no differences between the osteopathy and placebo groups in defecation patterns, eating habits, sleep, mood and crying (19). We were unable to find any studies that examined the efficacy of chiropractic, or that investigated the risk of injury in a systematic manner.

Osteopathy and manual therapy have many common features, but there are key differences in the comparisons made by the efficacy studies in this review. Osteopathy was compared with placebo (19). Manual therapy was compared with conservative measures, but whereas Haugen *et al.* (17) designed their study to test the efficacy of manual therapeutic manipulation in isolation, Cabrera-Martos *et al.* (18) examined a more complex manual therapeutic approach. These differences mean that we were unable to combine the results in a meta-analysis, but the studies should nevertheless be considered in association with one another. One possible interpretation is that conventional physiotherapy and osteopathy both promote symmetrical development, but the specific manipulation or impulse technique used is not crucial to the end result.

The results we report in this article are consistent with other reviews on the topic (26–28), but some uncertainty remains over the conclusions. The small number of studies and participants means that there is uncertainty as to the true effect estimates. It could be argued that studies with alternative designs should also have been included. However, as infantile asymmetries often resolve spontaneously, uncontrolled studies would not provide reliable answers with respect to the issue of efficacy (29, 30).

There is anecdotal evidence that manipulation treatment has led to adverse events in adult patients (31). Adverse events have also been reported in children, but in the context of manipulation treatment performed on patients who had received an inadequate work-up (24, 25). Effective diagnosis and work-up are important to avoid delays in treatment and to prevent exacerbation of underlying disease (32, 33).

Irrespective of the risk of serious adverse events, it is important to consider whether treatment may cause the patient discomfort. In a survey of Norwegian manual therapists who treat infants, 25 % reported that children often begin to cry in association with treatment (3). It is ethically problematic to offer treatment of unproven efficacy to children, especially if the treatment entails discomfort (34). However, it is also important to be aware that not all infants taken to manual

therapists, chiropractors or osteopaths due to asymmetries receive treatment with manipulation techniques (3). Half of Norwegian manual therapists report, for example, that they never manipulate the spine when treating infants, and that parental guidance is by far the most commonly used measure (3).

Data from Statistics Norway and the Norwegian Health Economics Administration show that approximately 8 % of all Norwegian infants are taken to manual therapists in their first year, often because of infantile asymmetry (3). This is in addition to treatments by chiropractors, osteopaths and other therapists. Why so many parents of infants seek help is unclear, but many may feel compelled to act if they perceive that their child differs from what is considered normal. A Finnish study showed that many concerns related to infantile asymmetries could probably be avoided if the parents receive good information and guidance when the child is born (35).

New parents are highly receptive to advice, and healthcare professionals should ensure that the advice given to parents, and the treatment given to children, is evidence-based and well-established. No causal relationship has been established between functional disorders of the neck and infantile asymmetries, and there is no reliable research to support the claim that untreated functional disorders of the neck give rise to complications and impairments later in life. This article, too, shows that the use of manipulation techniques to treat infantile asymmetries lacks evidence from research. Since the current evidence base consists of a small number of studies with few participants, it is difficult to draw firm conclusions, but our findings do not support the use of manipulation techniques in the treatment of infantile asymmetry.

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