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Systematic Review of Effects of Manual Therapy in Infants with Kinetic Imbalance due to Suboccipital Strain (KISS) Syndrome

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Abstract: The goal of this systematic literature review was to establish the effects of manual therapy, chiropractic, or osteopathic treatment of the kinetic imbalance due to suboccipital strain (KISS) syndrome in infants with positional preference, plagiocephaly, and colic. We searched PubMed, Embase, and the Cochrane Library for articles on the effects of manual therapy, chiropractic, and osteopathy for the KISS syndrome. In addition, experts in the field of manual medicine and osteopathy were asked to provide relevant articles, and the bibliography in a textbook of manual therapy for children was hand-searched for additional references to the syndrome. This search strategy located no clinical trials that evaluated the effects of manual therapy or osteopathy on either the KISS syndrome or its symptoms. However, pooled analysis of two randomized clinical trials on the effects of chiropractic in infantile colic showed no statistically significant difference between active and control treatments. In addition, we found that 22% of infants showed short episodes of apnea during manual therapy of the cervical spine and that one case has been described in which apnea after Vojta therapy resulted in death. Given the absence of evidence of beneficial effects of spinal manipulation in infants and in view of its potential risks, we conclude that manual therapy, chiropractic, and osteopathy should not be used in infants with the KISS-syndrome outside the context of randomized double-blind controlled trials.

Key Words: KISS Syndrome, Manual Therapy, Chiropractic, Osteopathy, Systematic Review

During the first year of life, approximately 8% of infants show positional preference of the head¹⁻⁴, which is associated with flattening of the skull (plagiocephaly) in more than half of the cases⁴. Other abnormalities such as torticollis, scoliosis, or limited hip abduction are also common in these infants⁴. When such symptoms are accompanied by excessive crying (colic), some practitioners have assumed a causal relationship with functional abnormalities in the suboccipital-high

cervical spine region. To describe this clinical entity, these clinicians coined the term “kinetic imbalance due to suboccipital strain (KISS) syndrome”^{5,6}.

In particular in Germany, Belgium, and the Netherlands, the KISS syndrome has received considerable attention in the lay press and on the Internet⁷. A group of manual physical therapists in the Netherlands has published a broad spectrum of possible symptoms and clinical associations to describe the KISS syndrome (Table 1). Spectacular improvements in symptoms have been reported after manual therapy⁶, chiropractic⁸, or osteopathic treatment of infants and toddlers with symptoms of the syndrome^{9,10}. Although techniques vary between therapists, “soft”, i.e., non-thrust, techniques are most commonly used to treat this condition.

Meanwhile, many medical physicians believe that the syndrome does not exist or that it constitutes a “fashionable disease”¹¹. Three years ago, however, the Dutch Associations for Pediatric Physical Therapy and

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Manual Therapy reached a consensus on joint treatment of infants with asymmetry and plagiocephaly¹², which seems to indicate that the KISS syndrome has entered the realm of mainstream medicine in Western Europe. Yet, uncertainty remains with regard to the beneficial and potential adverse effects of spinal manipulations in these young children.

In an attempt to evaluate the scientific basis of the KISS syndrome and its treatment by spinal manipulation, we performed a systematic review of the effects of manual, chiropractic, and osteopathic therapy on infants with symptoms of the syndrome. This study was designed to answer the following question: Are spinal manipulations, as performed by manual physical therapists, chiropractors, or osteopaths, effective in alleviating signs and symptoms of the KISS syndrome in infants, and what, if any, are the undesired side effects of this treatment?

Methods

We designed a search strategy to retrieve clinical trials and other studies on the effects of these treatment modalities. Because KISS syndrome is not a Medical

Subject Heading (MeSH) term, we searched Medline (1966-2004) through PubMed by retrieving articles with KISS syndrome as a text word in title, abstract, and key words, or with Biedermann H as the author because this practitioner is viewed as one of the founding fathers of the KISS concept⁶. We also searched the Embase and the Cochrane Library databases in a comparable way. Several websites^{7,12} on the KISS syndrome and a textbook on manual therapy in children¹³ were hand-searched for additional relevant studies. In addition, we approached the Dutch Association for Manual Therapy and several expert therapists in the Netherlands and Germany and asked them to provide further studies, if available.

When it became obvious that this search strategy was yielding no studies for systematic review, we performed an additional Medline (1966-2004) search for studies on the effects of spinal manipulation in infants with colic (MeSH terms “colic” and “spinal manipulation”) limited to “All infants (0-23 months)”, and studies on the effects of spinal manipulation in young children in general (MeSH term “spinal manipulation”) limited to “All infants (0-23 months)”. We performed a hand-search of the reference lists of the studies retrieved with this search strategy for additional references. No specific

Table 1. Excerpts from a waiting room flyer for parents on possible signs and symptoms of KISS syndrome in infants (reproduced from a flyer downloadable in the Dutch language from www.ewmm.net; accessed Dec 1, 2004)

<p>The KISS syndrome is possible if one or more of these are present:</p> <ul style="list-style-type: none"> • Vacuum or forceps delivery • Wrong positioning of infant in the womb • Cesarean section, multiple pregnancy • Prolonged labor, very rapid delivery • Poor sucking and swallowing • Vomiting (reflux) • Poor sleeping pattern • Hyperextended sleep posture • Flattening of the skull • Oblique skull with bald spot on one side • Curved spine (scoliosis) • Developmental dysplasia of the hip • Reduced movement of one body side • Very early tendency to stand up • Does not like being cuddled • Skips crawling phase • Early walking – as early as 7 months • Stumbles and falls often • Restless and hyperactive behavior • Delay in language development • Headaches • Poor concentration skills • Fall on the head (also after infancy) 	<p>Early characteristics</p> <ul style="list-style-type: none"> - Head turned obliquely to one side - Asymmetry of face - Excessive crying, poor sleeping - Curved spine (scoliosis) - Strong tendency to overextension - Asymmetric hip development - Asymmetric movement of arms and legs - Problems swallowing, vomiting - Crying when dressing or undressing <p>Late characteristics</p> <p>This is a list of possible characteristics at later age:</p> <ul style="list-style-type: none"> - Child does not want to crawl; early standing and walking - Poor and slow development of gross and fine motor skills - Equilibrium problems, poor posture - Stumbling often, falling with poor correction - Delayed language development - Fatigue, anger, temper tantrums - “Seems” to need little sleep - Wakes up with headache - Headache and poor concentration at school - Forgets tasks and assignments - Insecure, restless, attention-seeking
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search strategy was used to locate articles on possible harm of spinal manipulation, but studies retrieved with the search strategy discussed above were appraised for data on harm.

Results

KISS syndrome in infants

The searches in Medline, Embase, and the Cochrane Library for studies on the KISS syndrome yielded no hits at all. We retrieved one article on KISS syndrome in Russian when searching for papers with Biedermann H as author but this study was not a clinical trial. Without any relevant clinical trial, a further analysis of the effects of spinal manipulation in infants with the KISS syndrome could not be performed.

Spinal manipulation in infants with colic

This search yielded two studies on chiropractic treatment for colic, both from Denmark^{14,15}. Table 2 provides further data on these studies. In both studies, the chiropractic treatment was described in general terms. Physical examination included motion palpation of the articulations of vertebral spine and pelvis. Those articulations found to be restricted in movement were manipulated/mobilized with specific light fingertip pressure. These manipulations were performed in repeated sessions until normal mobility was attained. The difference in parent-reported outcomes between the two studies was rather striking. In the study where parents were aware which treatment their child had received, a statistically significant effect of chiropractic spinal manipulation on signs and symptoms was found¹⁴. In the other study, however, where parents and examiners were blinded with respect to the treatment received by their children, crying behavior improved to a comparable degree in the groups receiving active

and sham chiropractic treatments¹⁵. After pooling of these methodologically heterogeneous studies, no effect of chiropractic treatment on crying behavior after 8 weeks was found (Table 2).

Spinal manipulation in infants

We were not able to locate any additional studies on spinal manipulation in infants.

Safety of spinal manipulation in infants

One fatality after spinal manipulation in an infant has been described. This case report¹⁶ described a 3-month old baby girl referred for asymmetric posture who had been treated with Vojta therapy. After one such treatment session, the baby developed reflex apnea, followed by massive and fatal brain edema¹⁶. The Vojta session involved pressure on the right ulnar humeral condyle and on the dorsum of the right upper leg with the infant in a prone position with her head rotated to the right. The intensity and velocity of the manipulations involved was not mentioned in the paper¹⁶. Immediately after the manipulation, the infant cried and sweated profusely; a few minutes later, she stopped breathing¹⁶. In a group of 199 infants treated with manual therapy (with the “soft” non-thrust manipulations of the cervical spine mentioned in the introduction as the most commonly applied technique in such infants), “vegetative reactions” were found in 54% of patients, including apnea of short duration (<10 sec) in 22% of cases¹⁷, profuse sweating in 8%, flushing in 49%, and bradycardia¹⁸ for up to 10 sec in 42%.

Discussion

This systematic review provided no evidence supporting spinal manipulation by manual physical therapists, chiropractors, or osteopaths, in infants with signs

Table 2. Results of two randomized controlled trials of the effects of chiropractic spinal manipulation on crying behavior in infants with colic (mean reduction in hours of crying per day, as recorded by parents in a crying diary, between start of therapy and follow-up at 8 weeks)

First author Year of publication	Chiropractic therapy		Control therapy		Between-group difference in hours of crying after 8 weeks (95% confidence)
	n	Reduction of crying hours (SD)	n	Reduction of crying hours (SD)	
Wiberg et al (1999) ¹⁴	25	2.7 (1.5)	15	1.0 (1.5)	1.7 (0.7 to 2.7)
Olafsdottir et al (2001) ¹⁵	42	2.0 (2.9)	32	2.3 (2.9)	-0.3 (-1.6 to 1.0)
<i>Pooled</i>	67	2.3 (2.4)	47	1.6 (2.5)	0.7 (-1.6 to 2.2)

- Control therapy consisted of dimethicone in the Wiberg et al¹⁴ study and of sham chiropractic treatment in the Olafsdottir et al¹⁵ study
- In the Wiberg et al¹⁴ study, parents were aware which treatment their child had received; in the Olafsdottir et al¹⁵ study, they were not.

and symptoms proposed to be indicative of the KISS syndrome. In fact, no clinical trials on the outcomes of manual therapy in infants and children were found at all. The pooled analysis of two studies examining the effects of chiropractic treatment in infants with colic showed no statistically significant effect of this treatment (Table 2).

We deliberately set out to design a sensitive electronic search strategy. It has been shown that medical databases such as the ones we searched have limited sensitivity in finding studies on manual therapy¹⁹. We did not have access, however, to more sensitive databases such as MANTIS (Manual Alternative and Natural Therapy Index System). Therefore, we limited our search not only to the available electronic databases but we also approached experts in the field in the Netherlands and Germany as well as professional associations and we hand-searched reference lists in retrieved articles and in a textbook written by one of the proponents of the syndrome¹³. As a result, we think it is unlikely that we missed any published clinical trial on the KISS syndrome in infants. However, publication bias (negative trials not having been published) cannot be excluded.

We deliberately limited our search to randomized controlled trials. We excluded uncontrolled observations of groups of patients, many of which are available, both in print and on the Internet^{7,13} because a considerable placebo-effect can be expected and because most signs and symptoms (such as plagiocephaly and excessive crying) hypothetically associated with the KISS syndrome show favorable natural histories without specific interventions^{4,20}. For example, this placebo effect was clearly present in the studies on chiropractic therapy for colic (Table 2).

Based on the veritable tidal wave of enthusiastic accounts in the lay press and on the Internet, it can be safely concluded that many parents of infants with KISS syndrome are convinced that their children's symptoms are considerably improved after spinal manipulation^{6,21-23}. It could be argued that such observations mirror data from randomized controlled trials in adults with shoulder and neck pain where significant treatment benefits of manual physical therapy have indeed been described²⁴⁻²⁹. There are two reasons, however, why the anecdotal success stories of spinal manipulation therapy of KISS syndrome in infants should be interpreted cautiously and perhaps skeptically. First, the syndrome is poorly defined (Table 1), and a well-described and commonly accepted pathophysiological mechanism is lacking. The most commonly proposed hypothesis is that birth trauma, in particular after prolonged labor or vacuum or forceps delivery, is responsible for the "kinetic imbalance" in the suboccipital-high cervical spine region in these infants⁶. However, beyond this proposed pathophysiological etiology, there is no scientific support for this hypothesis. In studies of infantile colic or

positional preference, no associations with prolonged labor or troublesome delivery have been found^{4,30}. Second, our systematic review shows a complete absence of randomized controlled clinical trials on the effects of spinal manipulation in infants with the KISS syndrome. The commonly heard view that manual therapy and other forms of spinal manipulation in infants with the syndrome have been shown to be effective in clinical practice has as yet not been scientifically validated, and it is certainly not supported by the results of this systematic review of the literature on this topic.

In medicine, it is not unusual to continue using a certain form of therapy, even in the absence of evidence from randomized controlled trials, if there are no doubts regarding the efficacy and especially the safety of the treatment. For example, even though controlled clinical trials on immobilization of bone fractures or on appendectomy in acute appendicitis are lacking, the usefulness of these procedures is obvious and doubted by no one. In our view, however, this line of reasoning is not applicable to spinal manipulation in infants with KISS syndrome. Because many signs and symptoms of the syndrome, including positional preference⁴, plagiocephaly², and colic³¹, tend to resolve spontaneously, it is unclear whether treatment is needed at all. Moreover, it is not unreasonable to assume there may be risks involved in spinal manipulation in young infants. Chiropractic treatment of the cervical spine in adults is associated with a small but significantly increased risk of stroke^{32,33}. During manual therapy of infants with KISS syndrome, vegetative reactions, bradycardia, tachycardia, and reflex apnea have been recorded in more than half of patients^{17,18}. Although the apnea was usually of short duration (<10 sec) and reversible¹⁷, this can be regarded as a potentially life-threatening adverse effect of treatment. In a well documented case of an infant with symptoms of KISS syndrome who died after Vojta therapy involving manual forces inducing regional cervical rotation, reflex apnea was identified as the probable cause of death¹⁶. Although the therapy applied in this particular case is different from the non-thrust mobilizing technique usually applied by manual physical therapists for the KISS syndrome, the profuse sweating and apnea observed in this case¹⁶ appear similar to some of the "vegetative reactions" observed in infants with the syndrome after such non-thrust manual therapy techniques^{17,18}.

Conclusion

This systematic review revealed no scientific evidence that spinal manipulation by manual physical therapists, osteopaths, or chiropractors is useful in infants with signs and symptoms of the proposed KISS syndrome. The evidence reviewed here indicates that chiropractic treatment of infants with colic is not useful.

Only rigorously designed studies can further delineate the underlying causes, pathophysiology, symptoms, signs, and appropriate treatment of the proposed KISS syndrome. The beneficial effects of spinal manipulation in infants with this syndrome, if any, and its safety should be evaluated in randomized, controlled, double-blind clinical trials. Until the results of such studies are available, we strongly advise against performing spinal manipulation in infants because of the potential risks involved, based on the results of this systematic review into the potential benefit and harm associated with this intervention.

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