

What are the characteristics of infants with KISS?

- An inter-tester reliability study of the manual therapy classification of infants with KISS

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Introduction

A significant group of Norwegian manual therapists have during the last three to four years, treated many babies with asymmetrical movement patterns and in particular babies with a rotated head position. Previously these children were treated by pediatric physical therapists with the diagnosis of torticollis, where it was assumed that the etiology behind this misalignment was dysfunction of the sternocleidomastoid muscle. The treatment was for most parts, in addition to stimulating interventions, passive stretches. Recent research is questioning the effect of stretches. In addition this has been, to some extent, a long lasting and extensive treatment. Over the last years manual therapists and manual doctors in Central-Europe have investigated whether the asymmetrical position in the infant may have another explanation than muscular dysfunction. Functional disturbance of the upper cervical joints as a reason for torticollis is a relatively new way of defining this symptom. Heinrich Biedermann has described this phenomenon, both with emphasis on evaluation- and treatment method (Biedermann, 2004; Biedermann, 2004). Meanwhile it's a fact that there is, as of today, no scientific documentation neither on diagnostic tools or treatment method of this group of patients. My wish has been to contribute to increased knowledge of which findings can provide indicators to if the infant needs manual therapy treatment for asymmetrical position of the head. The term KISS (from German «Kopfgelenk Induzierte Symmetrie Störung») is here used to describe a «Kinematic Imbalance due to Suboccipital strain».

Despite extensive searches in the literature I have not come across any prevalence studies of KISS. The occurrence of other related dysfunctions have also been investigated. This is mainly favorite positions of the head and cranial asymmetries (Boere-Boonekamp & al, 2005; van Vlimmeren, Helders, van Adrichem, & Engelbert, 2006) and congenital muscular torticollis (Heidal, Dancke, & Martinsen, 2001). Meanwhile KISS has been introduced as a term both in Central-Europe and Norway without this being described in scientific articles internationally.

What is KISS?

Biedermann divide these children into two groups, KISS I and KISS II.

In brief KISS I is recognized by a «fixated» lateral flexed neck, while KISS II is recognized by the head fixated in a retro flexion (extension).

KISS I - fixed lateral flexion



Typical findings here are (Biedermann, 2004)

- C-scoliosis of the neck and body, meaning there is a lateral flexion to one side and a rotation of the head to the opposite side
- Cranial asymmetry
- Unilateral microsomia (tiny abdomen)
- Asymmetry of the gluteal area
- Asymmetrical use of the extremities
- Unilateral delayed motor development

KISS II – fixated retro flexion



Typical findings (Biedermann, 2004)

- Hyper-extension of the neck, in particular during sleep, the head is frequently rotated to one side
- Flattening of the occipital area, most often asymmetrical
- Elevated shoulders
- Fixated supination of the arms
- Inability to extend the body in prone position
- Orofacial muscular hypotonia
- Problems with breastfeeding on one side

Reasons

Reasons for the functional disturbance of the upper cervical joint is not yet clear. Robby Sacher (Sacher, 2004a) states that both prenatal and perinatal risk factors can be an explanation. As an example intrauterine space relations, the delivery procedure, type of delivery, facilities used such as vacuum extraction and cesarean sections (Sacher, 2004a; Philippi et al., 2006). We will not outline these reasons any further in this article.

Motor development

Examining spontaneous motor skills is a natural part of the evaluation when assessing an infant's asymmetrical movement pattern. It is expected, for a three month old infant with normal motor development, to maintain a stable supine position with legs flexed and the head kept steady at midline position. The infant should take an interest in good eye contact and may maintain looking at a toy with the head rotating to the right and left without the body following. Both arms grasp for toys. By the end of the second trimester the infant will be able to grasp its feet without rolling on to the side. In prone position you will look for symmetrical arm support, center of gravity will gradually move downward towards the sternum and the pelvis and the head may move freely to either side and without reclinuation or lateral flexion of the back or the neck (Mork, 1989; Schjetlein & Mossige, 1997).

During the first year of living the brain will, which is developing rapidly, gradually control (inhibit) the primitive reflexes and gradually be released by the postural reflexes. The postural reflexes give the basis for controlling the automatic movements, such as head control, adjusting the body in balancing and coordinating movements (Goddard, 2002).

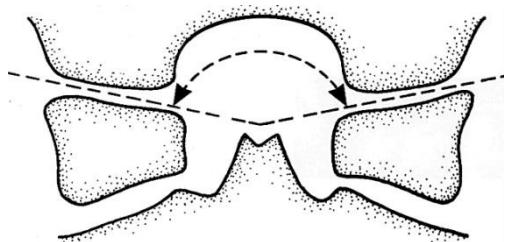
The most central position reflexes in this context are the tonic labyrinthine reflex and the tonic neck reflex. The tonic labyrinthine reflex contributes to maintaining a constant head position in the room. This is essential in maintaining the body's balance. The labyrinthine reflex is activated by the equilibrium organ in the inner ear. The neck reflexes contribute to maintaining a constant body position in the room (Brodal, 2004). These two reflexes together are a functional unit which depends on each other and together controls the development of posture and body movement. These reflexes develop during the first three months of life (Brodal, 1997).

The tonic labyrinthine reflex and the tonic neck reflex are controlled from the upper cervical joints and we consider that a dysfunction here may have major consequences for the children (Biedermann, 2004). The congenital, primitive and postural reflexes are also essential in motor development. The receptors for these reflexes are located in the upper cervical joints, which further emphasizes the need for normal function here.

Biomechanics

It is shown that the functions in the upper cervical joints are somewhat different in the infant compared to the adult, which refer to before and after vertical positioning (Biedermann, 2005) (Sacher, 2004b).

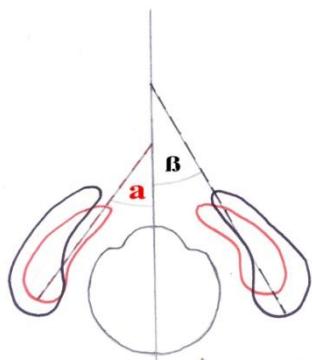
Biomechanical character of an infant:



(Biedermann, 2005)

Fig 1 Atlanto-occipital view with a frontal angle

In infants you will see a flattening of the atlanto-occipital joint in the frontal plane compared to the adult (Fig 1). The occipital condyles' position related to the sagittal plane has a more open angle in infants than in adults (Fig 2) (Biedermann, 2005). According to Biedermann the biomechanical consequence is that atlas becomes too mobile, or «floating» in infants. This may lead to a blockage of the upper cervical joint. In addition the head of the infant is relatively large and heavy. Since the muscular control of the connection between the head and the body is not fully developed, Biedermann states that the sub-occipital area is vulnerable the first months of living.



(Biedermann, 2005)

Fig 2 Atlanto-occipital view in the sagittal plane

- a) In infants (28°)
- b) In adults (35°)

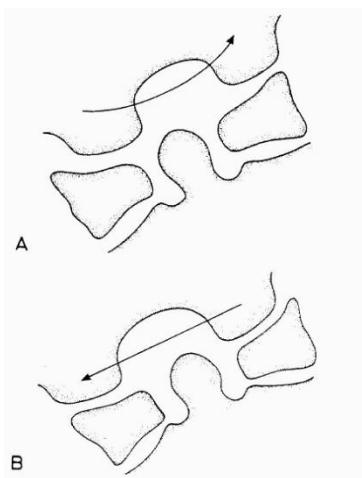


Fig 3 Lateral flexion, A) Adults B) Infants

Due to the shape of the joint surfaces, atlas will also move opposite in the frontal plane compared to in the adult (Biedermann, 2005) (Fig 3). In lateral flexion of the head in adults,

atlas glides towards the concave side of the flexion, due to the shape of the joint surface. On x-rays of infants we have seen the opposite happening, meaning that atlas has a tendency to glide towards the convex side of the flexion. The importance of this evidence is if we should treat these children and in which direction we ought to correct a possible malalignment.

Differential diagnostics

Several diagnoses can have similar symptoms as KISS. This may be dysplasia, anomalies, tumors, defects, pareses or problems related to vision and hearing. The list is long, something that tells you that you must pay attention to the fact that asymmetry may have many other reasons other than muscular or arthrogenic dysfunctions.

Purpose and problem

The main purpose of this project is to examine inter-tester reliability amongst manual therapists in classifying infants to have KISS or not have KISS. The criterion upon which the classification is based on is the evaluation of spontaneous motor skills, anti-gravitation side-tilt- test and segmental mobilization of the upper cervical joint. Good reliability will create a basis for further studies on the subject. Since spontaneous motor skills are such an essential part of the evaluation, a pediatric physical therapist was included in the project for this specific evaluation.

Even if there, as of today, are no clearly defined criteria in order to diagnose/classify infants with KISS, the project had to begin with describing how the manual therapists classify KISS in the clinic. The goal has been to describe today's tests used in the clinic with emphasis on purpose and how they are performed. The goal for the future is to be able to specify a standard for how manual therapists classify this group of patients, which again can form the basis for further studies on interventions in this area.

Problem:

- 1) How are manual therapists classifying the infant to have KISS or be healthy in the clinic?
- 2) Test-retest (inter-tester reliability) of the manual therapy evaluation of the infant with KISS.
 - How good is the reliability between manual therapists?
 - To what degree do the pediatric physical therapists agree in the evaluation of what is considered normal and abnormal in motor development?

Data and method

This study is an inter-tester reliability study of the evaluation of selected children, of which half are diagnosed by the «manual therapist 1» (MT1) to have KISS and half as healthy (Hennekens CH, 1987). In this study MT1 was represented by two experienced manual therapists from a consensus group, which was responsible for selecting the children. MT 1 does in this context represent the golden standard for the result. My own evaluation (MT2) of the children were done together with an experienced pediatric physical therapist (PPT) where she exclusively observed the children looking at the spontaneous motor skills.

Selection

22 infants, 11 healthy (assessed by MT 1) and 11 infants with KISS (classified by MT 1) were included in the study. These were evaluated in regards to spontaneous motor skills, side-tilt-test and segmental mobility in the upper cervical area. All of the children were recruited from Oslo with a geographically even distribution of the entire city.

Healthy infants were recruited from the community health center and from other colleagues in Oslo. KISS-infants were recruited by manual therapist colleagues in Oslo.

Inclusion criteria

- Infants between 3 to 6 months of age, corrected age in premature born.
- Both healthy infants and KISS infants (half-and-half).
- The parents had to agree to have the child evaluated by the therapist representing the consensus group, and as soon as possible thereafter seeing me for the same evaluation.

I selected the age group three to six months. Studies have shown that newborn frequently have a position preference with a rotation of the head to the right in supine (Geerdink, Hopkins, & Hoeksma, 1994). Geerdink has in the same study also shown that from 12 weeks of age the infant prefer to keep the head at midline and that this probably is related to improved head control, this made it natural for me to look at infants which have passed the age for congenital positional preference of the head.

Exclusion criteria

- Suspecting serious pathology.
- Down's syndrome.
- Infants with intermittent torticollis.
- Infants which recently have been treated by manual therapist or chiropractor.

Consensus group

The criteria to be able to classify which infants have KISS and which are healthy, were defined in the introduction. To help me in this I established a consensus group. The group consisted of three experienced manual therapists which have treated KISS infants over several years, in addition to myself. At selecting clinical tests and measures we wanted tests and criteria that are used in the clinic today. The goal for the study was to reflect clinical practice today, and to which extent you can call this standardized practice. We chose tests which we considered the most central in order to classify infants to have KISS or be healthy.

Clinical tests defined by the consensus group

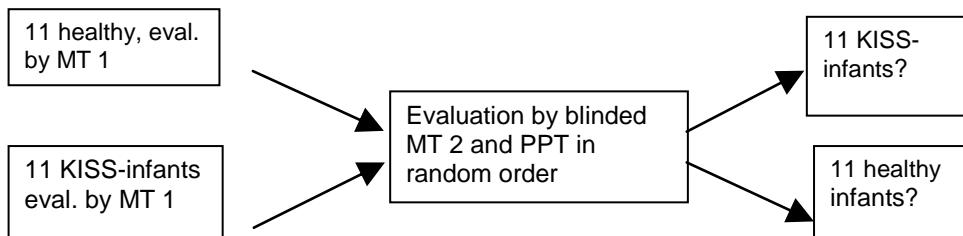
- Spontaneous motor skills
- Anti-gravitation activity in side-tilt-test
- Passive rotation of the neck
- Segmental mobility tests of the upper cervical joints, lateral flexion and flexion.

The decision whether the infant became classified with KISS or to be healthy was based on an overall evaluation of all the tests.

In addition a patient history form was passed out to all the parents for completion. This information did not take part in the classification of KISS/healthy, but were used after the evaluation in order to assess own compliance between our evaluation and the parent's.

A closer description of the performance of selected tests, and detailed procedures, can be found on www.manuellterapi.no > For members

Briefly summarized the procedure can be displayed like in this diagram:



Ethics

This project became accepted of REK Øst- Regional committee for medical research and Norsk samfunnsvitenskapelige datatjeneste AS- for collection and storing of patient personalia.

Results

There are no difference between the two groups related to demographical data (age, gender, birth weight and born at due date or before/after)

The therapists classification the infants

Table I: Briefly summarized the results can be displayed like this:

	MT1	MT2	PPT
Healthy/normal	11	12	16
KISS/not normal	11	10	6

- In 21 of the 22 infants there is full compliance between MT1 and MT2.
- In 10 of the 11 Kiss infants there is full compliance between MT1 and MT2.
- In 17 of the 22 infants there is full compliance between all therapists.
- In all the children who got diagnosed with KISS the parents reported on the patient history form that the infants had an asymmetrical movement pattern.
- In 14 of the infants the parents reported an asymmetrical movement pattern, that means also in the three infants classified as healthy by MT1 (according to the «golden standard» which is MT1)
- Cranial asymmetry was unanimously reported on all the children with KISS, but also in three of the healthy infants.

- Two of the children with the KISS diagnosis were delivered by a cesarean section, and three of those diagnosed as healthy were delivered by a cesarean section.

Kappa value, agreement by percent and P-value

Data was entered into SPSS, Survival manual (Pallant, 2007b). In Table II there are results of all the infants put together. When we pulled out the 19 that were referred by one of the MT1, the values became more or less the same. Here the Kappa value became 0,890 between MT1 and MT2, consequently no essential difference. The same is for the other values, which have only minor differences. A Kappa value of .8 or above represents very high agreement, in this case between MT1 and MT2. Between MT1 and PPT and MT2 and PPT there is moderate agreement, even though somewhat improved between MT2 and PPT than between MT1 and PPT (0.62 vs 0.55) (Pallant, 2007a). Equivalent findings can be seen in agreement by percent and the P-value.

Table II:

	Kappa value	% agreement	P =
MT 1 vs MT 2	0,91	95,45 %	<0,000
MT 2 vs PPT	0,62	81,8 %	<0,002
MT 1 vs PPT	0,55	77,7%	<0,004

Discussion and conclusion

The results display a very high agreement and good Kappa value between MT1 and MT2, which means that there is a very high inter-tester reliability between the manual therapists in this survey. Testing of the segmental mobility in adults with cervicogenic headache has shown similar test-retest reliability (Hanten W P & more, 2002). The size of the selection group is too small to be able to make definite conclusions, but this is a pilot study which can give a solid base for similar studies with a larger number of infants.

The agreement between MT1 and PPT is somewhat lower than between MT2 and PPT. There was an agreement in 17 of the 22 infants. Between MT2 and PPT there was an agreement in 18 of 22 infants. With a closer look MT1, MT2 and PPT agreed of all the healthy while PPT evaluated more infants to be healthy than MT1 and MT2. There can be many reasons for the different outcomes. Probably the manual therapists and pediatric physical therapists have different criteria for what is defined as KISS is what is defined as healthy, or what are considered to be the boundaries within normal motor development (Mork & Amdam, 1989). There may be vague boundaries, something which may provide room for different interpretation of what we observe. Another issue is that PPT in this survey exclusively observed the infants looking at spontaneous motor skills, something which does not reflect common clinical practice for pediatric physical therapists. She touched the children only to move them from supine to prone. In the manual therapy evaluation there were performed clinical tests on the neck, in addition to observation. The manual therapist therefore obtained more detailed information since the evaluation was more extensive.

Was the selection of the tests successful? And were the grading satisfactory? The main purpose of this study is to look at the reliability of our clinical tests. I have therefore chosen to describe the performance of the tests as precise as possible, so it will be possible for

others to repeat. In this study there is a high degree of agreement which points towards that the tests have properties that make them adequate in testing segmental mobility in the upper cervical joints of an infant. It remains to be seen if the validity of these tests are satisfactory. Do we test what we aim at testing and does segmental mobility have anything to do with asymmetry? These are questions that this study does not answer.

The consensus group is limited to people chosen by the author. Maybe the outcome would have been different if the randomization process had been different and had included other colleagues with other opinions of which tests are relevant and/or other ways to perform and interpret the tests. This is left to be exposed in further studies.

In such a study it is essential to consider validity and reliability in a method study. In this evaluation there was a very high degree of reliability on the test-retest between two manual therapists, since there was an agreement in 21 of 22 infants. But did we find the right ill? It is of today premature to say anything about this since we do not have any methods for telling whether or not we chose valid tests, or criteria, for what I wanted to measure.

Internal validity will in this context be affected by many factors. Were there any systematic mistakes during the data collection, did the children change between the first and second evaluation, the communication with the parents, crying and disturbance in the children which were evaluated and filling out of the forms, are all factors that could influence the outcome.

External validity is decided by whether the selection is representative, or not, the size of the selection and confounding (Bakketeig & Magnus, 1993). Factors as selection and design will lead to a confined generalization which poses a threat to external validity (Domholdt, 2000). The selection in this study is recruited from Oslo and may therefore not automatically represent the country as a whole. The way that the children were recruited will probably influence the outcome in the shape of selection unevenness (Bakketeig a al., 1993). Like a pilot study I believe that the selection is representative for what was the goal in this project. We were able to try out the method and this gives a basis for further research. Another issue can be the size of the selection, in particular related to if it is possible to conclude based on the results. Within the frames of a time limited master study it became hard to evaluate more infants than what we did. The extent of this study will therefore be too small for me to be able to generalize the results. The hope is that my results may form the basis for larger studies at a later point, with similar design. A confounder can be a variable which occurs systematically, but here we have not given that any considerations (Bakketeig and more, 1993). A possible confounder can be that you today, out of fear of SIDS, discourage parents to let their children sleep on their stomach. This is revealed in an information letter from Statens Helsetilsyn in 1997 (The Government's Health Survey, 1997) which strongly recommends this to the parents. This means that all infants today sleep on their backs and there is therefore a greater possibility that they will favor a position and acquire cranial asymmetry. From this study it is not possible to draw a conclusion of whether this has affected the results.

Conclusion

This is a pilot study on inter-tester reliability between two manual therapists and between manual therapists and a pediatric physical therapist with the aim to classify infants to have KISS or not to have KISS. This study shows a very high reliability between the manual therapists and moderate reliability between the manual therapists and the pediatric physical therapist. This indicates that the clinical tests that are used in this study works well in obtaining the purpose and can be used in further studies. The study describes in addition clinical practice for manual therapy classification of the infant with KISS or healthy and describes what the signs are for infants with KISS. The pediatric physical therapist's evaluation is not described here. There are no scientific studies found on diagnosing or treating KISS. The goal of this study will form the basis for further studies on this professional subject.

Summary:

Background: Manual therapy intervention on infants with asymmetry has increased the last few years. Earlier infants which were diagnosed with a misalignment of the head were considered to have congenital muscular torticollis, but in recent years there has been a hypothesis that this misalignment may be due to a functional disturbance of the upper cervical joint; KISS. There is no scientific documentation on either diagnostic tools or treatment methods of this patient group.

Purpose and problem: The main purpose of the project was to examine the inter-tester reliability between two manual therapists in particular in classifying infants to that have KISS or not have KISS. The study also looked at inter-tester reliability between manual therapists and between manual therapists and a pediatric physical therapist, where the pediatric physical therapist evaluated the spontaneous motor skills to be normal/disturbed. Another goal was to describe clinical tests used in clinical practice today with thought on purpose and how they are performed.

Data and method: In cooperation with the consensus group it was decided upon that the criteria for classifying KISS/healthy should be the evaluation of spontaneous motor skills, anti-gravitation side-tilt-test, passive rotation of the neck and two segmental tests on the upper cervical joints. 22 infants ages three–six months were included, of which 11 were classified to have KISS and 11 to be healthy.

Results: Inter-tester reliability between manual therapists got a Kappa value of 0,91 and an P-value of 0,000 (very high agreement). There was a lower reliability between the manual therapists and the pediatric physical therapist with a Kappa value of 0,62 and an P-value of 0,002 (moderate agreement).

Conclusion: This is the first study in this area and the study is a pilot study. Based on the criteria which were used to be able classify the infants to KISS/healthy there was very high reliability between the manual therapists in particular in classifying the infants to KISS/healthy, while the pediatric physical therapist evaluated more infants to be within the frame of normal motor development. Further studies must be done to examine the validity of the diagnostic criteria used in this study.

Keywords: Infant, asymmetry, functional disturbance of the upper cervical joints, classification, inter-tester reliability.

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