ABSTRACT

Aim: The purpose of these studies was to undertake a survey of functional and cosmetic status in children treated for congenital muscular torticollis (CMT), to examine validity and reliability of the Muscle Function Scale (MFS), to find reference values for rotation and lateral flexion of the neck and muscle function of the lateral flexors in the neck for the normally developing infant, to investigate if infants with CMT are at higher risk of achieving the early motor milestones later compared to a control group of healthy infants and to investigate if treatment duration is affected when stretching is carried out by an experienced physiotherapist compared to parents.
Methods: Range of motion (ROM) in neck rotation was measured with an arthrodial protractor. Lateral flexion was measured with the infant/child lying in supine on a big protractor. Muscle function of the lateral flexor muscles of the neck was measured with MFS, which was also tested for validity by a panel of experts. Physiotherapists and students tested intra-rater and inter-rater reliability of the MFS using photos. The presence of asymmetry of the face, posture and lateral band were observed and estimated according to a scoring sheet in study I. In study IV and V craniofacial asymmetry and head posture was assessed with the visual scale "severity assessment for plagiocephaly". Motor development was assessed with Alberta Infant Motor Scale. A questionnaire about time spent in prone when awake and sleep position was used. Infants with CMT were randomized to stretching treatment by physiotherapist or parent in study V.
Results: The majority of the children who had received earlier treatment for torticollis attained an overall excellent/good status and the most notable findings were remaining craniofacial asymmetry and asymmetry in muscle function. The MFS had high inter-rater and intra-rater reliability, weighted Kappa and intraclass correlation both >0.9. Reference values for the mean ROM in neck rotation in healthy infants were in mean 110° with SD 6,2° and a range of 100°-120°. In lateral flexion the mean ROM was 70° with SD 2,2° and a range of 65°-75°. Infants of two months of age had the mean muscle function score of 1, which increased to 3-4 at the age of ten months. Difference in scores on the left and right side were rare. Multiple regression analysis showed that infants in the CMT group had a significantly lower score at AIMS compared to the control group at two ($p=0.03$) and six months of age ($p=0.05$). Infants who spent ≥ three times daily in a prone position when awake, had significantly higher scores at AIMS than infants who spent less time in prone at two ($p=0.001$), six ($p <0.001$) and ten months of age ($p <0.001$). When stretching treatment was performed by an experienced physiotherapist the time to achieve satisfactory ROM in both rotation and lateral flexion was significantly ($P<0.01$) shorter compared to the parents group. Symmetrical head posture was achieved earlier ($P=0.05$) in the physiotherapist group than in the parent group
Conclusion: Most children with CMT had an overall excellent/good status at follow up after physiotherapy treatment and the most notable findings were remaining craniofacial asymmetry and asymmetry in muscle function. The MFS was found to be valid and reliable. Infants under one year of age have good ROM in rotation and lateral flexion of the neck. Infants with CMT seem to be at higher risk of achieving the early motor milestones late compared with a healthy control group. However time spent in prone position seems to have a positive influence on this. Infants with CMT gained full ROM and symmetric head posture earlier when treated by an experienced physiotherapist compared to parents. Nevertheless parents can achieve a good result within a couple of months.

Keywords: Craniofacial asymmetry, rotation, lateral flexion, muscle function, infants, reference values, torticollis, early motor milestones, stretching treatment, physiotherapy.

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SAMMANFATTNING

Syfte: Huvudsyftet var att göra en kartläggning av funktionellt och kosmetiskt status hos barn som tidigare har behandlats för congenital muscular torticollis (CMT). Att utvärdera validitet och reliabilitet hos Muscle Function Scale (MFS), att ta fram referensvärden för rotation och lateralflexion i nacken och för muskel funktion i nackens lateralflexorer hos spädbarn, att undersöka om barn med CMT har ökad risk för att uppnå motoriska milstolpar senare jämfört med en kontrollgrupp, samt att undersöka hur behandlingstiden påverkas då en erfaren sjukgymnast eller föräldrarna genomför stretching.
Metod: Range of Motion (ROM) i rotation mättes med en arthrodial protractor och vid mätning av lateralflexion låg barnen på en stor gradskiva. Muskel funktion bedömdes med MFS. En expertpanel bedömde validitet för MFS, sjukgymnaster och sjukgymnaststudenter testade inter- och intrabedömar reliabilitet med hjälp av fotografier. Ansiktsasymmetri, huvudhållning och muskulär sträng bedömdes med en poängtabell i studie I. I studie IV och V bedömdes craniofacial asymmetri och huvudhållning med en visuell skala "severity assessment for plagiocephaly". Referensvärden togs fram genom mätning/bedömning av spädbarn utan några kända åkommor. Motorisk utveckling bedömdes med Alberta Infant Motor Scale och ett kort frågeformulär användes för vaken tid på mage och sovposition. Barn med CMT randomiserades till stretchingbehandling av sjukgymnast eller föräldrar.
Resultat: De flesta barn tidigare behandlade för CMT hade ett utmärkt/bra status och de mest påtagliga fynden var kvarstående craniofacial asymmetri och asymmetri i muskelfunktion i nackens lateralflexorer. MFS hade god validitet och hög intrabedömar och interbedömar reliabilitet, weighted Kappa och intracalass correlation var båda >0.9. Referensvärden för spädbarns nackar: i rotation var medelvärdet 110° med SD 6,2° och range 100°-120°. I lateralflexion var medelvärdet 70° med SD 2,2° och range 65°-75°. Vid två månaders ålder hade barnen i medel 1 poäng på MFS och poängen ökade successivt till 3-4 vid tio månaders ålder. Analys med multiple regression visade att barn med CMT hade signifikant lägre poäng på AIMS jämfört med en kontrollgrupp vid två månader (p=0.03) och sex månaders ålder (p=0.05). Spädbarn som tillbringade ≥ 3 tillfällen per dag på mage när de var vakna hade signifikant högre poäng på AIMS än barn som tillbringade mindre tid på mage vid två (p=0.001), sex (p <0.001) och tio månaders ålder (p <0.001). När stretchingbehandling genomfördes av erfaren sjukgymnast var behandlingstiden för att uppnå tillfredställande ROM i både rotation och lateralflexion signifikant kortare (P<0.01) än för föräldrar. De barn som fick stretching av sjukgymnast istället för av föräldrar uppnådde också symmetrisk huvudhållning tidigare (P=0.05).
Slutsats: De flesta barn behandlade för CMT hade ett utmärkt/bra status och det mest påtagliga fynden var kvarstående craniofacial asymmetri och asymmetri i muskelfunktion. MFS har god validitet och reliabilitet. Spädbarn har en god passiv ROM i nackens rotation och lateralflexion. Spädbarn utan kända åkommor har symmetrisk funktion i nackens lateralflexorer enligt MFS. Barn med CMT tycks ha en ökad risk för att uppnå tidiga motoriska milstolpar senare men vaken tid i magläge tycks ha större betydelse för den motoriska utvecklingen. Barn med CMT uppnår tillfredställand ROM och symmetrisk huvudhållning tidigare när en erfaren sjukgymnast genomför stretchingbehandlingen. Föräldrar klarar dock att få ett tillfredställande resultat inom några månader.

Sökord: Craniofacial asymmetri, rotation, lateralflexion, muskelfunktion, spädbarn, referensvärden, torticollis, tidiga motoriska milstolpar, stretching, sjukgymnastik.

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LIST OF PAPERS

I Öhman A, Beckung E. Functional and cosmetic status in children treated for congenital muscular torticollis as infants. Advances in Physiotherapy. 2005;7:135-140.

II Öhman A. Nilsson S. Beckung E. Validity and reliability of the Muscle Function Scale, aimed to assess the lateral flexors of the neck in infants. Phys Theory Pract. 2008 Accepted.

III Öhman A. Beckung E. Reference values of motion and muscle function of the neck - in infants. Ped Phys Ther. 2008;20(1):53-58.

IV Öhman A. Nilsson S, Lagerkvist A-L, Beckung E. Are infants with torticollis at risk of having a delay in achieving early motor milestones compared with a control group of healthy infants? Dev Med Child Neur. 2008 Accepted.

V Öhman A, Nilsson S, Beckung E. Stretching treatment for infants with Congenital Muscular Torticollis physiotherapist or parents, the treatment dilemma. A randomized, controlled trial. Submitted.

CONTENTS

LIST OF ABBREVATIONS

AIMS	Alberta Infant Motor Scale
AROM	Active Range Of Motion
CMT	Congenital Muscular Torticollis
DP	Deformational Plagiocephaly
ICC	Intraclass Correlation Coefficient
LAR	Labyrinthine Righting reflex
MFS	Muscle Function Scale
MT	Muscular Torticollis
ORR	Optical Righting Reflex
PROM	Passive range of motion
PT	Postural Torticollis
ROM	Range Of Motion
SCM	Sternocleidomastoideus
SD	Standard Deviation
SIDS	Sudden Infant Death Syndrome
SMT	Sternomastoid Tumor

INTRODUCTION

Congenital muscular torticollis

Congenital muscular torticollis (CMT) is the third most common congenital musculoskeletal anomaly in infants next to congenital hip dysplasia and clubfoot. The reported incidence in the world is 0,4-1,9 % (1) however a recent study indicates that it might be higher (2). CMT is a result of shortening or excessive contraction of the sternocleidomastoid (SCM) muscle often with limited range of motion (ROM) in both rotation and lateral flexion. The head is typically tilted in lateral flexion towards the affected SCM in the frontal plane and rotated towards the opposite side in the transversal plane (figure 1) (1,2,3,4,5). After birth a painless sternomastoid tumor (SMT) that consists of fibrous tissue may be present, which in most cases will disappear within some months (6,7,8,9).

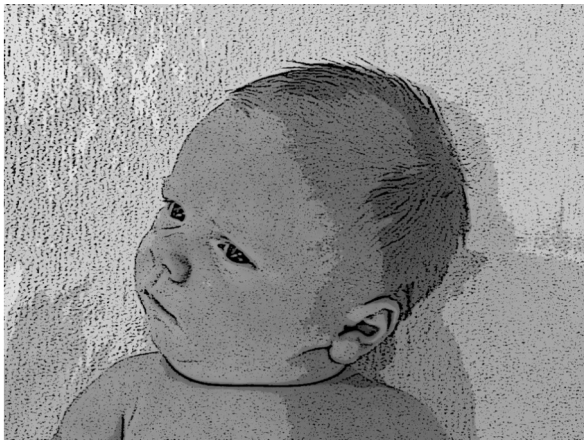

Figure 1. Infant with left sided CMT.
Typically tilting to the left and with rotation to the opposite side.

For many children with CMT there is an imbalance in muscle function around the neck, it has been found that the lateral head righting on the contralateral side is weakened compared to the affected side (1,10,11). Skull asymmetry i.e. deformational plagiocephaly with posterior flattening on the opposite side of the torticollis is common (12,13,14,15). Facial asymmetry occurs in the presence of prolonged uncorrected head tilt (7,16).

Associated findings

There is a preponderance of first-borns among infants with CMT (3,6,10,11,17,18,19). Breech presentation and other complications at birth are also more common than in the normal population. Breech presentation occurs in about 3-4% of all deliveries and it decreases with

advancing gestational age, at <28 gestations weeks the incidence is 35 % (20) and at term the incidence is 2-3% (20,21). Breech presentation for children with CMT is reported to be 17-46 % (3,6,10,11,18,19,22,23,24,25). Many reports in the literature have confirmed a consistent relationship between CMT and dysplasia of the hip (7,10,11,22,23,26). The incidence of dysplasia of the hip is approximately 1% of newborns (27) and hip dislocation in the unscreened populations is estimated to be 0.1-0,2 % in children of European origin (28,29,30,31). The reported coexistence rate of CMT and dysplasia of the hip is 4,1 % - 29 % (1,7,26, 32). Tien reported that dysplasia of the hip was found only on the ipsilateral side of the torticollis. However none of the patients with postural torticollis (PT) in this study were found to have dysplasia of the hip (26). Von Heideken et al found that boys were 4.97 times more likely than girls to have both dysplasia of the hip and CMT (32).

Sternocleidomastoideus muscle

The SCM is a large muscle that passes obliquely across the side of the neck (figure 2). It arises from the sternum and clavicle from two heads. The two heads are separated from each other at their origins by a triangular space, but then gradually blend, below the middle of the neck, into a thick, rounded muscle. This is inserted, via a strong tendon, into the lateral surface of the mastoid process, from its apex to its superior border, and also via a thin aponeurosis into the lateral half of the superior nuchal line of the occipital bone (33).

Figure 2. The sternocleidomastoideus muscle.

Histological studies and magnetic resonance imaging (MRI) shows muscle atrophy and interstitial fibrosis in the CMT muscle (34). The normal ultrasound presents as a hypoechoic

mass with echogenic lines, indicating muscle fascicles running throughout its length, CMT muscles tend to be more hyperechogenic (34,35). For infants with CMT the SCM muscle can be classified by ultrasonography into four types: I/ a fibrotic mass in the involved muscle, II/ diffuse fibrosis mixing with the normal muscle, III/ without normal muscle in the involved muscle, IV/ a fibrotic cord in the involved muscle (8,36). Compared to type I, children with type IV are at a significantly higher risk of needing surgery (8,36). Lin and Chou found that both the sternal and clavicular heads were involved in 76 % of their patients and only the sternal head in 24 %. No isolated fibrotic change was found in the clavicular head alone by Lin et al 1997 or by Cheng et al 2000 (35,37). The fibrotic muscle is unable to maintain growth as compared with the unaffected side. This causes shortening of the SCM on the affected side with limited ROM and head tilt as a result (38,39).

Sternomastoid tumor (SMT)

SMT is a benign palpable, firm, fibrous, nontender mass (figure 3) measuring from 1-3 cm and frequently found in the lower third of the SCM muscle (7,9,40,41). The incidence is 0,4 % in all newborns and infants (9). It is often diagnosed when the children are two to four weeks old and after reaching its maximum size the mass gradually disappears and in some of the children the muscle becomes fibrotic (5,41,42). The presence of SMT is not always associated with reduction in neck ROM (1,6).

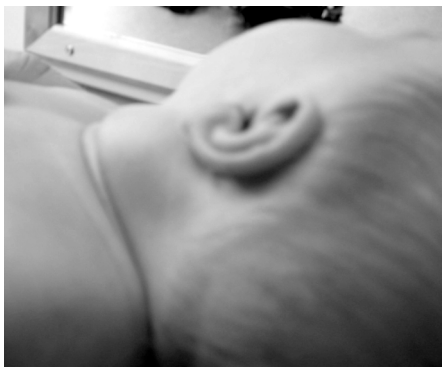

Figure 3. Infant with SMT in left SCM

Holloway in 1931 excised five SMT and Chandler et al in 1944 excised 26 SMT and neither of them found evidence of a prehistory of haematoma (4). The presence of SMT affects not only the size of the muscle, but its echo structure (35,36). The echo texture is more hyperechoic than the healthy contralateral or the unaffected adjacent muscles, and in the larger tumors the normal fascicular lines are disrupted (35,36). For infants with more severe

rotational deficits there is a significantly higher proportion of hyper echogenicity and abnormal appearance (35).

Aetiology

The cause of CMT has been speculated on since 1670. Genetics, birth trauma, ischemia and intrauterine malposition have all been considered (4). Sippel (1924) and Isigkeit (1931) noted that CMT could be detected in the uterus, as their radiological evidence indicated that the condition existed before delivery (4). Cadaver dissections and injection studies has defined the SMC muscle compartment (42). According to Ho et al and Do et al a primiparous mother has a smaller uterus and malposition of the foetus is not unexpected (3,34). Do et al says that this could lead to more difficult and traumatic deliveries (34). Chen et al screened 1021 newborn infants for CMT with sonography and found that the infants with CMT were statistically significantly longer and heavier than the normative group (43). In the past haematoma from birth trauma was often suggested as a cause of CMT. This theory has little support because hemosiderin has not been found on ultrasonography or in pathological specimens of excised masses (37,38). In clinic many infants are observed with a mandibular hypoplasia on the same side as the affected SCM muscle (figure 4). This indicates that the infant may have been in a malposition in uterus. Stellwagen found that the incidence of torticollis was higher when the mother reported the baby to be stuck in a position for more than six weeks relative to a shorter length of time or not at all, this supports the theory of malposition in uterus (2). In Ho et al study neonatal records for infants with CMT showed a presence of mandibular hypoplasia at birth in 59 % of the infants and all of them were noted on the affected side (3).

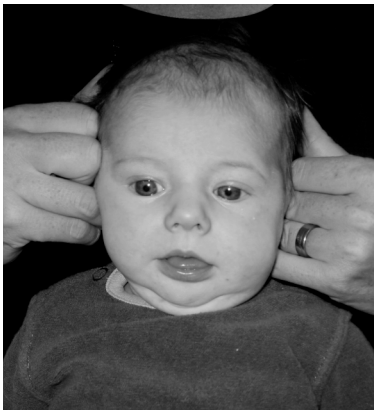

Figure 4. Asymmetry; infant with a smaller cheek on right side.

It seems to be a common knowledge among those who study CMT that the number of infants with CMT has increased since the "back to sleep campaign" (12,13). In the very young infants an early preference of head orientation in conjunction with long periods in a supine position and short periods in prone lying whilst not varying the head position, may combine to produce a tightness of the neck musculature on one side or a laxness on the other resulting in torticollis (12,14,44). If a muscle remains in a somewhat stretched position it tends to weaken whilst those that remain in a somewhat shortened position tend to be tighten and become stronger than their opponents (45). Even if the muscles are only slightly shortened or stretched, muscular balance can be upset if they remain in this position continuously or for a long period of time (45). As early as 1898 Heller carried out experiments on dogs, which showed that muscle ultimately shortens when its points of attachment are permanently brought closer together (39).

Differential diagnosis

Pathological processes that disturb the normal pathways controlling head and neck position can cause torticollis (46,47). Differential diagnosis ranges from innocuous abnormalities to potentially life-threatening tumors (46). In rare cases torticollis can be the initial clinical presentation of tumours of the posterior fossa and cervical spine (48).
More common non muscular causes may include skeletal abnormalities such as Klippel-Feil or neurological disorders, ocular torticollis, inflammatory illness and atlantoaxial rotatory subluxation etc (46). According to Kiwak over 80 causes of torticollis have been reported (47). One very rare differential diagnosis is muscle aplasia, described only in a few cases (49).

Classification

Torticollis can be divided into three groups: The sternomastoid group (SMT) including children with a sternomastoid tumour, Muscular torticollis (MT) which comprises children with tightness of the sternocleidomastoid but with no clinical tumour and Postural torticollis (PT) a group of children with all clinical features of torticollis but with no demonstrable tightness or tumour of the muscle (1,18,41) (table 1). The classification describes the condition at the time of the assessment; the infant may have been classified differently if assessed at an earlier or later date.

Table 1. Classification into torticollis group.

Torticollis group	Sternomastoid tumour	Limited range of motion	Tilt and/or rotation of the head
SMT	Yes	Yes	Yes
MT	No	Yes	Yes
PT	No	No	Yes

Range of motion in the cervical spine

The paediatric cervical spine approaches the adult configuration at eight years of age. The atlas, axis and sub axial spine each have unique patterns of ossification (50,51). General agreement exists among investigators regarding an age-related effect on the ROM of the extremity joints of newborns, infants and young children up to about two years of age. Mean values for these age groups differ by more than two standard deviations from adult mean values (52). Children tend to be very flexible, and it is natural that they lose some of this flexibility when their strength develops, as they grow older (45). Youdas et al found that both rotation and lateral flexion in the neck are significantly associated with age. In active rotation for children aged 11 to 19 years the mean was about 75°, it was rare with active ROM (AROM) of more than 80° after the age of twenty years (53). Arbogast et al found that children aged 3 to 5 years had an AROM in rotation of about 40-85° (mean about 70°) (54). Normally the passive ROM (PROM) is slightly greater than the AROM as each joint has a small amount of movement available that is not under voluntary control (53). With each 10-year change in age, Youdas et al believe that both genders lose approximately 3° in both rotation and lateral flexion of the neck (53). As AROM is dependent on the subject's muscle strength and coordination (52) it is preferable to use PROM when measuring infants. Infants with CMT have limited movement in rotation to the affected side and in lateral flexion to the opposite side. Measuring ROM is an essential part of the assessment for children with CMT as it gives information about the progress of the condition.

Deformational plagiocephaly

Deformational plagiocephaly (DP) is a condition in which the infant's head are deformed as a result of prenatal or postnatal external moulding forces to the growing cranium (55,56). The natural history of DP is likely to have existed for centuries although at a lower rate than present (13). Comparing the periods 1990 to 1992 with 1993 to 1994 Argenta et al found a dramatic increase of positional head deformity and found that all the affected infants were

supine sleepers (57). At the end of the last century the incidence of PT was estimated to 1.7 % (58). In a cross-sectional study published 2002 the incidence of cranial asymmetry in healthy newborns was 13 % (44). In a prospective cohort study performed year 2004-2005, 6.1 % of healthy newborn had DP and only 40 % of these infants had DP present at seven weeks of age. In the same study, 21 % of the healthy infants without DP at birth had developed DP at seven weeks of age (59). The increasing incidence is likely to be related to the "back to sleep campaign" (13,14,15). Infants with CMT prefer to rotate their head to the opposite side of the CMT; with this side preference there is a high risk of developing plagiocephaly. If viewed from the top of the infants' head the typical DP that forms a parallelogram will be observed. In addition to the usually unilateral flattening of the occipital area, there may be ipsilateral frontal and parietal bossing (figure 5), cheekbone prominence and anterior ear displacement ipsilateral to the flattened occiput (12,13,15,60,61,62).

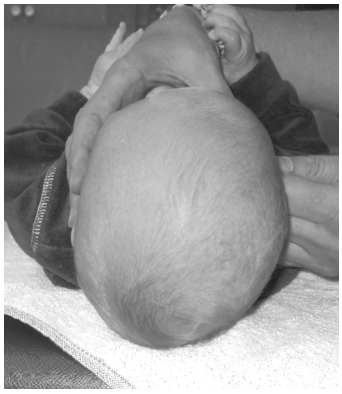

Figure 5. Infant with deformational plagiocephaly on the right side, posterior flattering and frontal and parietal bossing.

The skull undergoes 85 % of its postnatal growth within the first year of life (12). The growth is most rapid during the first six months of life, specially the first three months; hence the plagiocephaly can develop very quickly in a newborn (63). Significant deformity may persist in some individuals into adolescence and minor craniofacial asymmetry can be detected in a significant number of adults (13). It is very important to distinguish the DP from the rare condition of synostosis. With synostosis surgery will be considered. Factors associated with increased risk for DP at seven weeks of age: male gender, first-born birth rank, positional preference when sleeping, tummy time when awake <3 times per day and slow achievement of motor milestones. (59). Van Vlimmeren found that early achievement of motor milestones was a protective factor for developing DP and they also found that DP at birth was not found to be a predictor for DP at seven weeks of age (59).

Posture

Head tilt

At birth, infants already have the capacity to righting the head from either full flexion or full extension when they are supported in an upright position. At about two months, the infant can sustain the head in midline in the frontal plane during supported sitting (64). By the end of the third or fourth postnatal month, the head in conjunction with organized trunk and lower extremity extension has largely perfected the maintenance of stable positioning in space (64). In good posture the bone and joints are in position to take the stress of weight and motion, and the musculature is firmly balanced to hold the body in place (45). Infants with CMT tilt their head in lateral flexion more or less to the affected side. Faulty posture means being in a position of poor alignment all or most of the time, and the result can be an adaptive shortening or stretching of muscles (45). A persistent head tilt can result in facial asymmetry and cause cosmetic abnormalities (7). If a child has tilted the head for a long time the asymmetry of the face become more noticeable when the head is held erect (4). Faulty posture may cause fatigue, muscular strain and in later stages pain (45).

Shoulder Elevation

Children with CMT commonly elevate the shoulder on the affected side when there is a limited ROM and/or imbalance in muscle function (Figure 6) (4,18,19,65,66). In two Swedish studies elevation of the shoulder on the side of the CMT was frequently found, no child was found to elevate the shoulder on the opposite side (18,19).

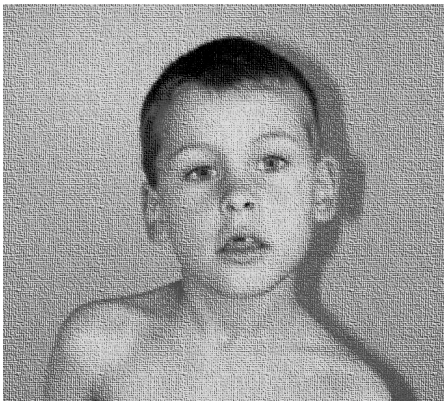

Figure 6. Boy with elevated shoulder on the affected side.

Also a lateral shift of the head towards the affected side can be seen (figure 7). According to Jones, most of the older children with CMT have a combination of these two features and either may be slightly predominant (4).

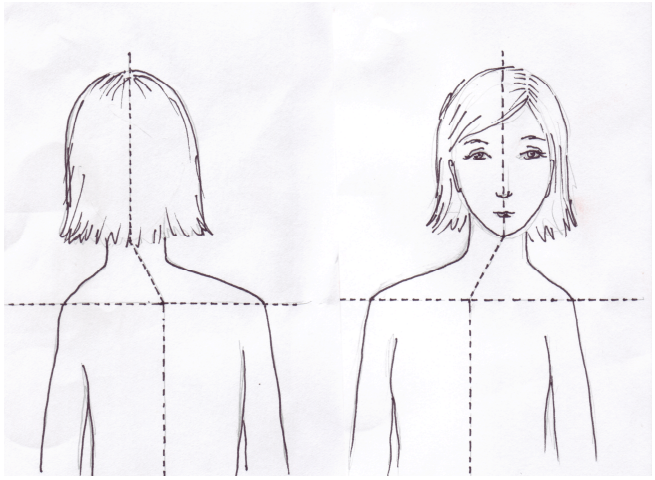

Figure 7. Lateral shift of the head towards the affected side.

Facial asymmetry

A varying degree of facial asymmetry is found predominantly before the treatment of CMT (figure 8) (5,25,67,68,69). Facial hemihypoplasia produces a flatter contour of the cheek and a lower orbital margin, its development, and disappearance after treatment, is related to torticollis (4,5,25,67,68,69). Minor degrees of asymmetry of the face and the cranium are universal. (4). Patients with multiple types of ocular torticollis, including face turns, show similar appearance of facial compression on the side of the torticollis, suggesting that the tilt or turn itself may cause the asymmetry (70). Improvement after the head is held erect is likely to continue as long as growth of the face and the skull continues (71). No improvement in facial asymmetry is to be expected after the age of 18-20 years (4,71).

Figure 8. Facial asymmetry.

Muscular imbalance in the lateral flexor muscles of the neck

Clinical examination commonly shows an imbalance in muscle function in the lateral flexor in the neck for infants with CMT (figure 9). Several authors have described that the lateral head righting on the contralateral side is weakened compared with the affected side (5,10,11,18,19). Several factors such as strength, endurance, power and length can affect the muscular function. Muscular function is essential to obtain a good posture, and it is important that the strength is well balanced to support bone and joints. The result of failure of the development of equilibrium can be a distortion of proper alignment and posture. When muscles are not balanced and when bones and joints are misaligned the body's structure must endure greater strain than intended (45).

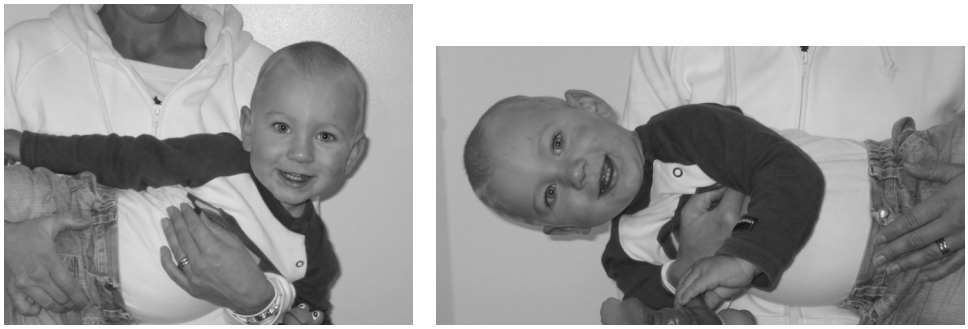

Figure 9. Infant with muscular imbalance, his right side shows a stronger muscle function than the left side.

Head righting response

Early in the development righting reflexes are expressed through muscular control; the movement against gravity will facilitate the righting reflex (72,73). The labyrinthine righting reflex (LAR) has an onset from birth to three months and the optic righting reflex (ORR) has an onset from birth to two months, both persist throughout life. When tilting an infant from a vertical to a horizontal position, there is a compensatory contraction of the neck muscles to keep the head in a vertical position (figure 10) (72,73,74). The LAR provides normal orientation of the head in space i.e. face vertical and mouth horizontal with gravity being the controlling factor. The ORR allows the head to be brought into normal position by using visual cues in the environment as a stimulus (73). As the infant develops and gains greater control of movement against gravity, the primitive reflexes decrease as the postural or automatic reactions appear (45).

Postural reflexes exist in muscle, the inner ear and in the cerebellum (75). For LAR the otolith organs of the inner ear stimulate the response, this reflex is coordinated in the

midbrain. For ORR the visual information keeps the head correctly orientated. This reflex involves the visual cortex as well as the brainstem (64).

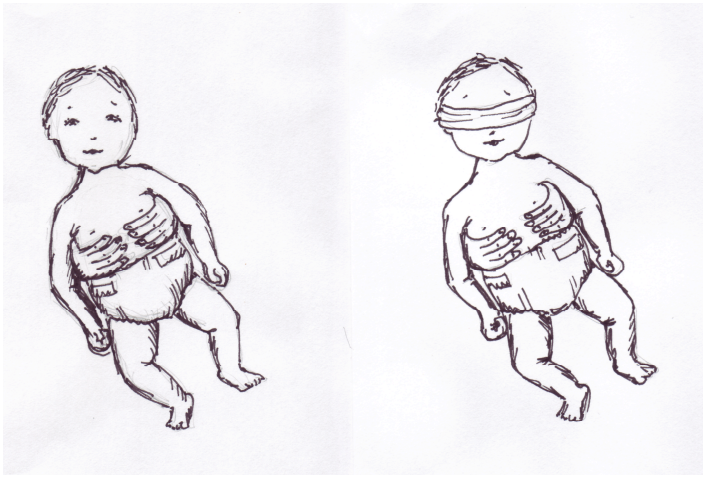

Figure 10. Head righting response in optical righting (ORR) and labyrinthine righting (LAR).

Infant motor development

A dynamical functional perspective on motor development currently drives most research on motor development (64). Dynamical systems theory emphasizes process and places neural maturation on an equal plane with other structures and processes that interact to promote motor development, such as muscle power, body mass, arousal, neural networks, motivation, and environmental forces (e.g. gravity and friction) (64). Postural control is a critical and integral part of all motor tasks and of motor development (72,73). Appropriate postural control is necessary for stability, balance and orientation (64,72,73,76). The systems that are considered of key importance for postural control are visual, vestibular and somatosensory. These systems produce orientation of the head in space and orientation of the body in relationship to the head and ground (72,73,76). This is essential for the maintenance of balance during movement and also ensures the appropriate body orientation for the specific task required (73,76). The newborns motor system is very immature; the first problem the infant faces is the impact of gravity (72,73,76). The ability to raise the head in prone position is seen as one of the first major motor achievements of the newborn (Figure 11) (64,72,73,76). It is suggested that the lack of head control is a result of both a lack of strength and a lack of organized muscle activity (73). The supine posture is important for promoting spontaneous activity in young infants (76). The centre of mass/gravity is proportionately higher in the first year of life and this requires large force generation and regulation by neck and upper trunk

musculature to counter the inertial forces created by displacements of the head (64,73). The change in the infant's centre of gravity during growth is of particular importance to balance (72,73,76). In order to sit unsupported, the infant must achieve appropriate head and trunk control (64,72,73,76). Maintaining balance in the upright position requires that the centre of gravity be kept over the supporting surface (73,76). Neuromuscular, musculoskeletal variables and the influence of experience are also important in motor development (73,76). The infant learns by repeating interesting or pleasing events that were originally experienced by chance (73,76). There can be considerable variability in development in children who ultimately achieve normal motor control (64,72,76). The major gross motor milestones of the first 12 to 18 months include achieving an indefinitely maintained upright head posture, attaining prone-on-elbows position, rolling from supine to prone, independent sitting, attaining hands-and-knees position, moving from sitting to four-point position and prone, creeping on hands and knees, pulling to stand, standing independently and walking independently (64,72). The motor milestones of creeping and walking are two of the most important in the infant's motor development. Once infants become mobile, they are more able to explore their environment (73,76). It is commonly assumed that there are no sex differences in motor ability in infancy (76,77,78).

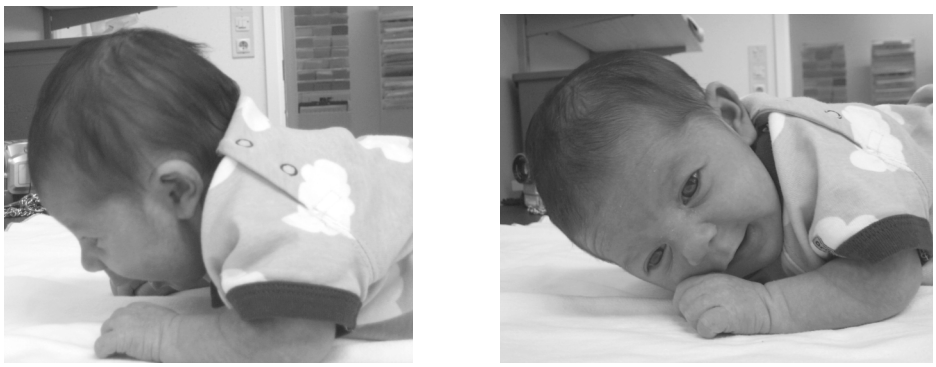

Figure 11. Head control, girl three weeks old with minor CMT.

Delayed infant motor development

One of the most common risk factors for developmental delay or disorders is premature birth (79). Preterm infants are often born with low muscle tone, or hypotonicity (79). For preterm infants there appear to be delays in the first two years before the infants "catch up" (79). For healthy full term infants the lack of experience in prone position can cause motor development to occur later (80,81,82,83,84,85,86,87). Also infants with torticollis seem to be at increased risk of late achievement of early motor milestones (88).

Alberta Infant Motor Scale (AIMS)

AIMS is a norm-referenced measure and consists of a 58-item observational assessment for infants, and assesses the infants' sequential development of motor milestones from birth to independent walking (89). AIMS measures spontaneous movements that reflect the quality of weight bearing, posture and antigravity skills in prone, supine, sitting and standing positions. The infant is assessed through observation with minimal handling and no arbitrary stimuli or facilitation. The infant will be given the opportunity to demonstrate the entire movement repertoire and there is no minimum or maximum number of trials for an infant to perform an item (89). No item shall be credited on the basis of development assumptions or parental reporting. The items are marked as "observed"(1 point) or "not observed" (0 point). Any items below the least mature item observed are credited with 1 point. The total AIMS score is a sum of the four positional scores. The AIMS is reliable and valid in discriminating the motor performance of normally developing infants from those at-risk and motor delayed infants, and for evaluating small changes in motor skills due to maturation (89,90,91,92). A ceiling effect has been found for AIMS at 12 months of age, and it is probably not suitable for use once an infant can lower her- or himself controllably from a standing position (93).

Paediatric physiotherapy in general

In paediatric physiotherapy research has become an important part and there is a responsibility of all members of the profession to deliver evidence-based treatment (64). Evidence-based practice involves "integration of best research with clinical expertise and patient values" (64,94,95). The guiding principle when working with children is the UN "Convention on the rights of the child" and the WHO definition of health "Health is a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity" (96). In paediatric physiotherapy the clinical decisions must be guided by the knowledge of the natural development of children (64). There is a need for physiotherapists to be familiar with the motor development and also with growth and development of the musculoskeletal system. The human skeleton undergoes an incredible amount of growth and change in the years between birth and skeletal maturity. What may be considered "normal" at a certain age can be decidedly abnormal at another (64). Knowledge of the milestones of cognitive development must be demonstrated in order to provide treatment in a stimulating and motivating environment and to take advantage of interactions between cognitive and motor development (64,73).

A useful model for the physiotherapy process (figure 12) is the International Classification of Functioning, Disability and Health (ICF) which has the overall aim to provide a scientific basis for understanding and studying health and health-related states, outcomes and determinates (64,73).

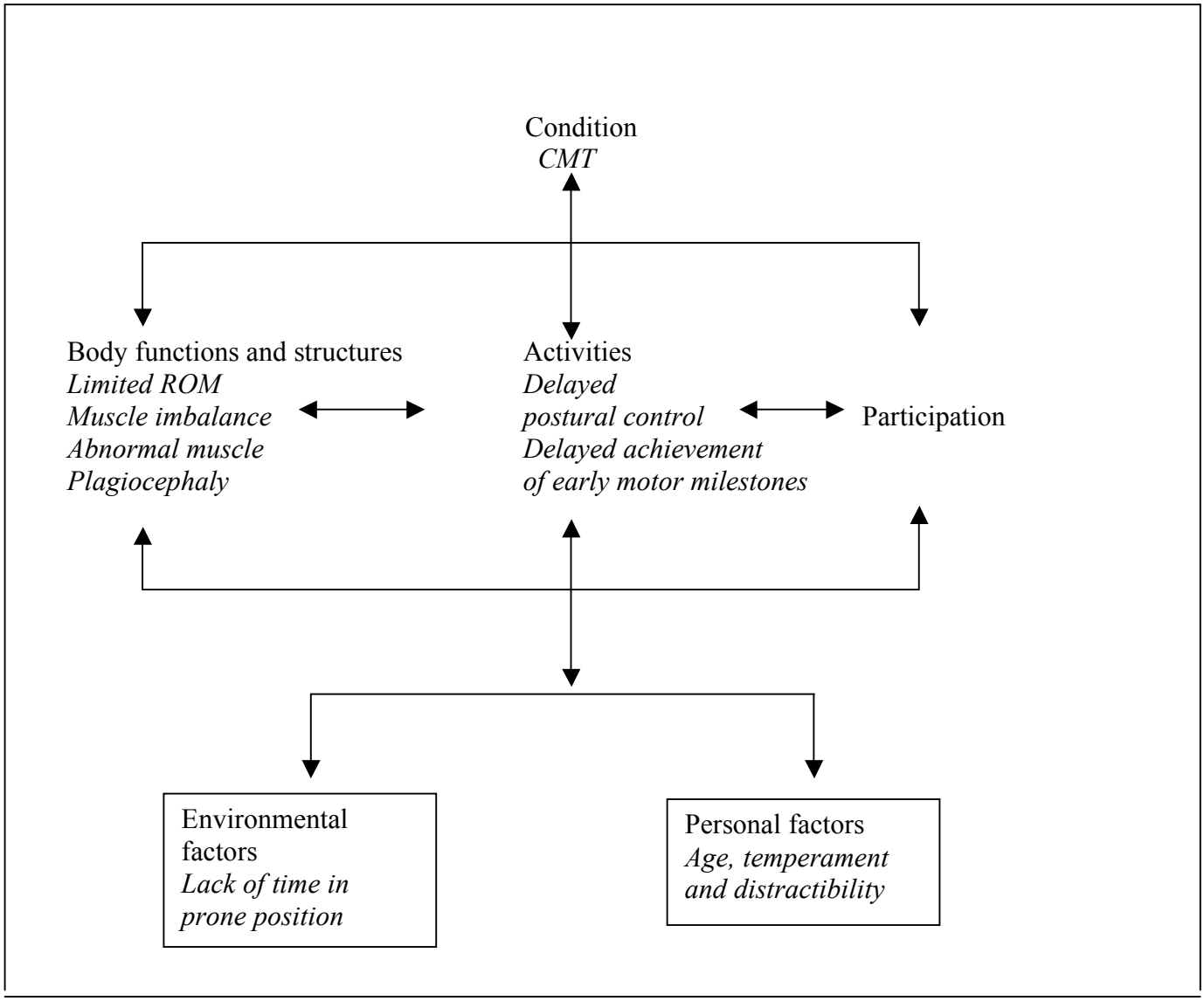

Figure 12. Typical findings that can occur: Congenital Muscular Torticollis (CMT) in the International Classification of Functioning, Disability and Health (ICF) model.

Treatment

Physiotherapy for CMT

Early treatment is essential and the primary goal in treatment is to prevent facial and skull deformities, limitation of neck movement, muscular imbalance and long-term posture change (10,34,67). Mostly it is the parents who perform the treatment with the paediatric physiotherapists teaching them a home programme with positioning, handling skills and stretching (10,11,17). The mean duration of treatment performed by parents is about 4-5 months (10,17). In studies there experienced physiotherapists performed all the stretching the mean duration of treatment was shorter (1,5). Stretching on the affected muscle is done in lateral flexion and rotation; the stretching can be done in different ways; for example with a combination of the both movements (figure 13). The hold of the stretch can be structured with a short hold and exact repeats (1) or individual adjusted treatment, with short or longer hold and repeats adjusted after what the infant accept. It is often necessary to distract the infant to get acceptance.

Figure 13. Example: of stretching exercise for left-sided CMT.

Examples of handling skills taught are how to carry the child to lengthen the sternocleidomastoid muscle, how to encourage prone playing and how to alter eating positions to diminish the side preference. Training of muscle function of the opposite side is also an essential part of the treatment even if it is often only briefly mentioned in publications (5,10,17). Muscle function training can be done both in handling and in specific exercises (figure 14).

Figure 14. Training of muscle function

When there is a marked tendency to turn the head to the side opposite the affected muscle, it may be possible to prevent the development of plagiocephali by putting the infant down to sleep on either side alternately, thus avoiding the supine position (4,15,44,57). When putting the infants to sleep in side lying they must be prevented from turning over in to a prone position because of the risk of sudden infant death syndrome (SIDS). Special pillows/wedges for this purpose can be used (figure 15)(15,97). According to a review article published 2008 there is not sufficient evidence to support the use of helmet therapy (98).

Figure 15. Special pillow/wedge for safe side lying

Since the "back to sleep campaign" the prone sleeping position decreased markedly and SIDS rates have decreased (13,14,15,29,57,58,99,100,101,102). Despite the increase of deformational plagiocephaly, the recommendation to use supine sleep position should be followed (13,99). Parents should be instructed to put the infant down to sleep in the supine position, altering positions of the head (left and right occiput) (13,66). When awake and being

observed, the infant should spend time in the prone position (3,12,13,14,44,97,100). The infant should spend minimal time in car seats (when not passenger in a vehicle) or other seating that maintains a supine position (12,97,100).

Surgery

In severe cases where there is no response to conservative treatment i.e. physiotherapy, the infant/child will be treated with surgery (1,103,104,105). For the majority of patients a uniform surgical method of distal unipolar open release with partial excision of the clavicular and the sternal heads of the sternomastoid muscle is carried out (1,25,67,68,103). For older children it may be necessary to carry out a bipolar release. After surgery there is an intensive programme of physiotherapy with stretching, strengthening and other exercises to stimulate a symmetric position of the head (1,103,105). The postoperative physiotherapy starts one to seven days after surgery (19,25,67,103,104). A postoperative brace is prescribed to help the child to maintain the corrected head position (figure 16) and is worn for one to several months (16,19). In some clinics an overcorrected position of the head is used (68,103). The best time for surgery is before five years of age but older children can also benefit (16,25,104,106).

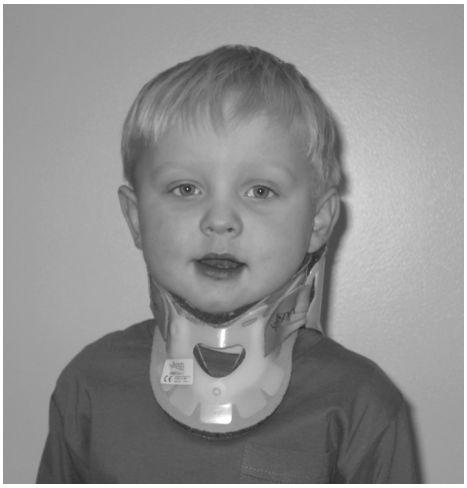

Figure 16. Postoperative brace treatment.

Prognosis

If CMT is left untreated the soft tissues may not grow in relation to the child's skeletal growth and this may result in significant permanent craniofacial deformities (4,104). Conservative treatment i.e. physiotherapy gives good results for about 95 % of the infants (5,10,22,41,103,107). The most important factors that will predict the outcome of manual stretching are the initial deficit in rotation of the neck, and the age of the patient at

presentation (10,41,108). The SMT group has the longest treatment period and the PT group the shortest (10,41). About 5 % of the infants with CMT are treated surgically.
The most common findings for persons who have had CMT are some degree of facial asymmetry and a tendency to tilt the head and/or elevate the shoulder (11,18,19,25,108,109,110). If CMT is untreated there may be a risk for cervical and facial cosmetic problems (3,67). The severity of cranial and facial deformity is correlated to age (4,16,110). Correction of facial asymmetry can occur as long as there is growth potential (62,110). A study in the Netherlands showed that the asymmetry in ROM and deformational plagiocephaly had not resolved in nearly one third of infant's with earlier signs of positional preference. These children were followed to the age of two to three years (111). It is unknown to what extent a spontaneous recovery for infants with CMT can be expected. However a recent study found that 16 % of the infants had a measurable torticollis at birth (2). This is much higher than the earlier reported incidence and it may indicate that there is a high rate of spontaneous recovery among those with mild torticollis.

Validity and Reliability

A measure is valid to the extent that it assesses what it is intended to measure (92).
To be reliable the measure must demonstrate the ability to differentiate among individuals and provide consistent values on repeated assessments. The reliability of the measurement and the competency of the therapists in performing them are essential to know when making clinical decision as this will indicate whether or not a particular measurement is of any value (112). Intra-rater reliability is parallel assessments of the same rater at different occasions when test-retest is used (92). Videotapes or photographs can be used to keep the attribute of interest stable (113,114). The Intraclass correlation coefficient (ICC) is often used to quantify relative reliability (112). Inter-rater reliability is parallel assessments by different raters, if all raters observe the same performance/videotape/photograph then the raters will be the only source of error (92).

Ordinal scales

Ordinal scales demonstrate an obvious order or hierarchy among response options; however the spacing among the responses is not viewed as being equal (92). Photographic and pictorial clinical grading scales have become available to practitioners. These grading scales are expected to be an improvement over verbal description scales because they add a degree of objectivity (115). The appropriate measure of agreement for ordinal data can be either

weighted kappa or ICC. A Kappa statistic of 0.81-1.00 is considered as almost perfect (116). An ICC of 0.7 is commonly used as a threshold of "sufficient reliability" (117).

AIMS

Study I

The aim of study I was to describe, in a cross sectional study the functional and cosmetic status in children earlier treated for CMT with regard to asymmetry of head position, ROM, muscle function i.e. strength/endurance and craniofacial asymmetry.

Study II

The aim of study II was to investigate validity and reliability of the Muscle Function Scale.

Study III

The aim of study III was to find reference values in normally developing infants for PROM in rotation and lateral flexion in the paediatric cervical spine and also to find reference values of muscle function in the lateral flexorers in the neck.

Study IV

The aim of study IV was to investigate if infants with CMT are at risk for late achievement of early motor milestones and to compare them with a control group of healthy infants. A second aim was to investigate if the time spent in prone position had influence on the early motor milestones.

Study V

The aim of study V was to investigate if there was any difference in treatment time when comparing stretching exercises carried out by an experienced physiotherapist or parents.

METHODS

Participants

Study I

All children treated for CMT between the years 1999 and 2001 at the Department of Physiotherapy at Queen Silvia Children's Hospital, Göteborg, Sweden were invited to participate in study I. Fifty-four children (72 %) between the ages 2-5 years participated (table 2).

Study II

Two groups which both comprised seven physiotherapists and two physiotherapy students tested inter-rater and intra-rater reliability.

Study III

Thirty-eight healthy infants participated during one or more occasions between the ages 2-10 months. They were recruited from the Child Health Centre in Göteborg, Sweden (table 2 and 3).

Study IV

A total of 122 infants between the ages 2 to 18 months participated in this study.
Eighty-two infants with CMT and 40 healthy infants participated as a control group (table 2 and 3).

Study V

Twenty infants with CMT and limited PROM participated (table 2).

A total of 190 children participated, the number of subjects in paper I, III, IV and V and the overlap between the studies are shown in table 4. In study II 13 physiotherapist and three students participated; one physiotherapist and one student participated in both groups.

Table 2. Gender and age of participants in study I, III, IV and V.

	Participants n	Female	Male	Age (months)
Study I	54	20	34	24 to 60
Study III	38	19	19	2 to 10
Study IV	122	53	69	2 to 18
Study V	20	10	10	0.6 to 18

Table 3. The number of infants who participated in the assessment using AIMS at one or three occasions before the age of 18 months. All infants who participated at six moths of age also participated also at ten and 18 months of age.

Group	Two months	Six months	Ten months	Infants who participated at one -four occasions	Infants who participated at three or four occasions (18 months included)	Infants who participated at all four occasions (18 months included)
CMT	25	54*	57	82	57	22
Control	35	39	39	40	38	35
Total	60	93	96	122	95	57

* for three of the infants that participated at two months of age there is data at ten months of age but missing data at six months of age.

Table 4. Distribution of subjects in the different papers.

Study	Subjects	
Paper I	54 children	54 new subjects
Paper III	38 infants	38 new subjects
Paper IV	122 infants	37 subjects from paper III 85 new subjects
Paper V	20 infants	7 subjects from paper IV 13 new subjects
Total		190 subjects

Dropouts

In study IV there were five infants with CMT who moved out of the area and for that reason could not fulfil the study**.** One infant in the control group participated only at two months of age in study IV as the parents found it had hard to fulfil the appointments. There were no dropouts in study I, III and V.

Measurement of range of motion

Study I

Neck rotation was measured with an arthrodial protractor. In study I the children were measured in a sitting position with a plumb line attached to the centre of the chin whilst the head was rotated along a vertical axis on a horizontal platform in front of the neck (figure 17) (18,19,109).

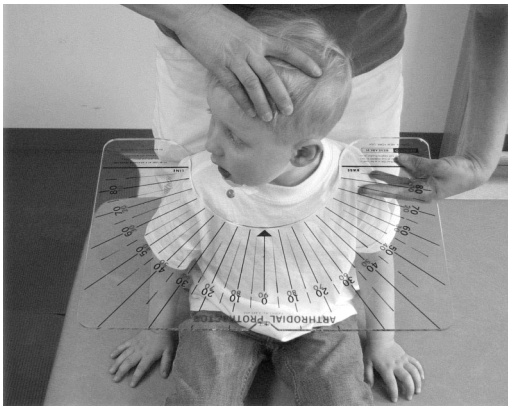

Figure 17. Measuring rotation with an arthrodial protractor in sitting.

Study III and V

In study III and V PROM of the neck rotation was measured with the infant in a supine position, with the shoulder stabilized and the head and neck supported by the examiner over the edge of the examination couch so that the neck was free to rotate and move in all directions (figure 18) (1,6,35). The interexaminer reliability correlation coefficient is 0.71 (41).

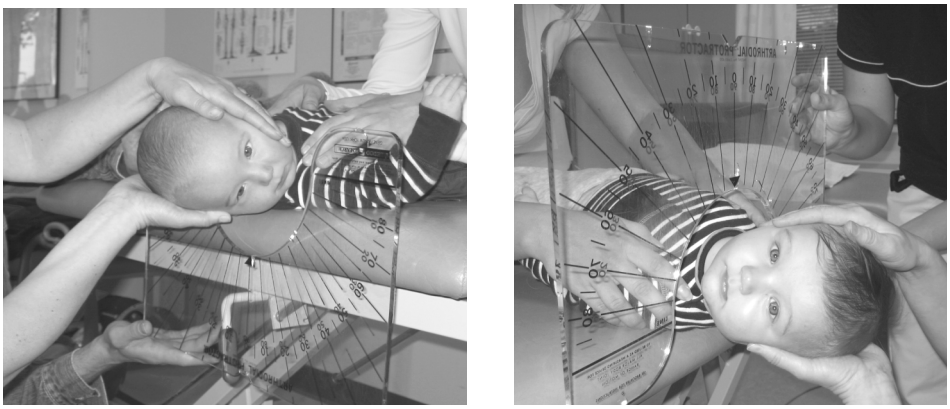

Figure 18. Measuring rotation with an arthrodial protractor in supine, to the right the protractor is held for rotation ≥90° and to the left the protractor held for <90°.

Study I, III and V

In study I, III and V lateral flexion was measured with the infant/child lying in supine on a big protractor with the shoulders stabilized (figure 19), (18,19). This method has been found to have high intra-rater reliability (118).

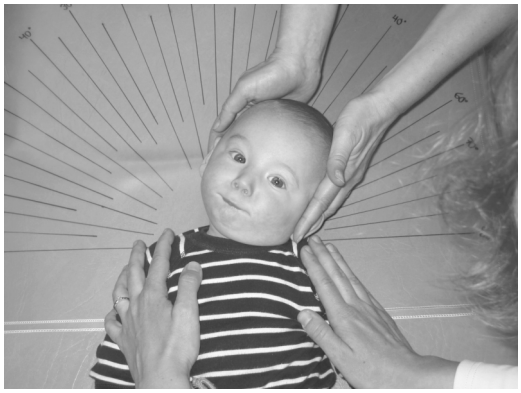

Figure 19. Measuring lateral flexion.

Estimates of craniofacial asymmetries, head tilt and lateral band

Study I, IV and V

In study I photographs were taken to gain a general impression of hypoplasia of one side of the face and to compare the sizes of the eyes, the horizontal levels of the eyes, ears and cheeks. A grid was used over the photographs when evaluating craniofacial asymmetry (119). Presence of asymmetry of the face, posture and lateral band were observed and estimated according to a scoring sheet used in several large studies (1,6,18,19,41,103,120). The scoring sheet is not yet tested for validity and reliability. In study IV and V assessment by severity scale from Cranialtech were used containing posterior flattening, forehead asymmetry, facial asymmetry and neck involvement (121). A multi-centre study is currently under way to determine the validity and reliability of this form (121).

Estimates of muscle function

Study I

Muscle function i.e. strength/endurance of the lateral flexor muscles of the neck was estimated with the child lying on the side lifting their head up (figure 21); time was counted in seconds (18,19). The youngest children who could not follow the instructions were held horizontally around the trunk without support for the head and the sides were compared. The head position was graded in a position over, at or under the horizontal plane.

Figure 21. Estimates of muscle function.

Study II, III and V

For study III and V muscle function of the lateral flexor muscles of the neck was estimated with MFS a five-degree scale with scores from 0 to 4 (appendix A). By holding the infant horizontally around the trunk without support for the head (figure 22), the degree of lateral head righting was estimated according to MFS. If the infant held their head below the horizontal line (can not hold the head against gravity) it scored 0, holding on the horizontal line scored 1, briefly above the horizontal line scored 2, high above the horizontal line scored 3 and very high above with their head almost in a vertical position scored 4.

Figure 22. Estimates of muscle function in an infant

In study II a panel of experts that provided insight, constructive criticism and advice discussed the MFS. The MFS was developed further to a six-degree scale (Appendix B) and reliability was tested with photographs. The scale was tested also with the scores described in degrees as suggested by one of the members in the panel of experts. Two groups which both comprised seven physiotherapists and two physiotherapy students tested inter-rater and intra-rater reliability. The MFS and the method of scoring were explained to the evaluators of reliability. They received a folder with 68 photos of infants with the body in a horizontal position and the

head in different positions. The infants on the photos were patients who had been treated for CMT at the clinic. The photos were selected so that all scores were represented. The evaluators were given the MFS with pictures of each score described mainly in words (group I) or in degrees (group II), written instructions on the method of scoring and a scoring sheet. No training of assessing with the MFS was given before the assessment. All evaluators did the assessment individually and were not allowed do discuss with anybody during the process. The assessment took approximately 15 to 30 minutes on each occasion. The same procedure was repeated with at least one week between assessments but the second time the photos were placed in a different order to reduce the possibility of memory recall. All photos were encoded with different codes on the two occasions.

Scoring for overall results

Study I

Overall results were graded according to a scoring system (1,6,18,19,41,120). The scoring sheet includes deficits in ROM in rotation and lateral flexion, craniofacial asymmetry, residual band, head tilt and a subjective assessment which included the parent's opinion of cosmetic and functional result (table 5). Clinical group, treatment date and time was collected from medical records.

Table 5. Scoring sheet for overall results by evaluation categories.

Overall results	Excellent	Good	Fair	Poor
Rotational deficits (degrees)	<5	6-10	11-15	>15
Side flexion deficits (degrees)	<5	6-10	11-15	>15
Craniofacial asymmetry	None-mild	Mild	Moderate	Severe
Residual band	None	Lateral	Lateral,cleido	Cleido,sternal
Head tilt	None	Mild	Moderate	Severe
Subjective assessment (cosmetics and functional)	Excellent	Good	Fair	Poor
Overall scores	16-18	12-15	6-11	<6

Assessment of early motor milestones

Study IV

The Alberta Infant Motor Scale (AIMS) was used to assess motor development at ages two, six, ten and eighteen months in study IV. The AIMS is a norm-referenced measure and consists of a 58-item observational assessment for infants. It assesses the infants' sequential development of motor milestones from birth to independent walking (89). The AIMS measures spontaneous movements, which reflect the quality of weight bearing, posture and antigravity skills in the prone, supine, sitting and standing positions (89).

STATISTICS

Study I

The Wilcoxon signed ranks test was used for calculation of differences between the affected and non affected side, in lateral flexion, rotation and endurance. The calculations were made in the exact degrees for ROM and in category for endurance. Spearman's rho was used for correlation of category data.

Study II

Weighted Kappa and intra-class correlation (CCI) was calculated for intra-rater and inter-rater reliability.

Study III

Mean, standard deviation (SD) and ranges were calculated for ROM in rotation and lateral flexion. Mean and range of the MFS scores were calculated for estimated muscle function.

Study IV

The Mann-Whitney test was used to compare AIMS percentile ranks. Suspected late motor development was tested using Fishers exact test. Differences in AIMS raw scores between CMT and controls were analyzed with ANCOVA using prone time awake, plagiocephaly, sleep position, gestation week, birth length and birth weight as covariates. The Wilcoxon signed-rank test was used to compare plagiocephaly at two and ten months of age.

Study V

Treatment time was logarithmed and analysed with ANCOVA with and without covariates age, lateral flexion and rotation.

Ethics

Informed consent was obtained from all parents. The local Ethics Committee at the Gothenburg University approved the studies. The measurements and estimates did not harm the children but use of discretion was necessary when assessing craniofacial asymmetry as there is a risk that children older than 2-3 years could become worried or affected in some way by the attention given to the face and skull deformity.

RESULTS

Study I

The majority of the children had an overall excellent/good status (table 6) and the most notable findings were craniofacial asymmetry and asymmetry in muscle function i.e. strength/endurance.

Table 6. The overall results of the survey of 54 children with congenital muscular torticollis.

Overall results	Excellent	Good	Fair	Poor
Rotational deficits (degrees)	48	5	1	0
Side flexion deficits (degrees)	48	4	2	0
Craniofacial asymmetry	29	11	13	1
Residual band	52	1	0	1
Head tilt	44	7	1	2
Subjective assessment (cosmetic and functional)	35	15	4	0
Overall scores	41	9	4	0

Clinical groups: there were 24 % in the SMT group, 61 % in the MT group and 11 % in the PT group. The age at the start of treatment was five days to three years (table 7). The duration of treatment varied with the longest being 24 months, (table 6). Also the number of attendances varied with the least being only once by one child. Fifty-two children were treated conservatively and 2 had surgery.

Table 7. Clinical groups of congenital muscular torticollis and the average age at the start of treatment and duration of treatment.

	SMT	MT	PT
Average age at the start of treatment	1,8 months (median 1,5 months)	2,8 months (median 2 months)	4,8 months (median 3,3 months)
Average duration of treatment	7,8 months (median 6 months)	5,3 months (median 4 months)	3,3 months (median 2,3 months)

Study II

The panel of experts found evidence of validity for the MFS. In the discussion the panel of experts found a discrepancy between the lower scores 0-1-2 and 3-4 and as a result of this the MFS was changed from five to six ordered scores to make the intervals between the scores more even. An additional version of the MFS described in degrees from the horizontal plane was made for testing purpose only. The Weighted Kappa and ICC was >0.9 for both inter-rater and intra-rater reliability of both versions.

Study III

Reference values for mean PROM in the neck rotation in healthy infants were 110° with SD 6,2° and range between 100°-120° (table 8). The measurements in rotation at two months of age were about 5° less than at the other ages. One infant had a difference of 5° between the right and left side in rotation. In lateral flexion the mean PROM was 70° with SD 2,2° and range between 65°-75° (table 8).

Table 8. Reference values for ROM in rotation and lateral flexion in 38 healthy infants under the age of one year. Measured on 2-4 occasions at the ages of 2, 4, 6 or 10 moths, in 37 of the infants.

Range of Motion (ROM)	Mean	Standard deviation (SD)	Number of measurements
Rotation	110°	6,2°	104
Lateral Flexion	70°	2,4°	112

Infants of two months of age had the mean muscle function score of 1 range 0 to 2. Muscle function score increased with age (table 9). There was no significant difference between genders. Two infants had a difference in muscle function between the right and left sides.

Table 9. Reference values of muscle function for healthy infants according to an ordinal muscle function scale 0-4. Head position estimated in relation to the horizontal line, 0=below, 1=on line, 2=slightly above, 3 high above and 4= very high above.

Muscle function score at age (month)	Mean	Range
Two	1.0	0-2
Four	2.6	1-4
Six	3.0	2-4
Ten	3.4	3-4

Study IV

Percentile rank of motor development was significantly lower for the CMT group at two ($p=0.02$) and six ($p<0.01$) months of age (Table 10) compared with the control group.
At two and six months of age there were significantly more infants from the CMT group at or below the tenth percentile according to the AIMS than from the control group (Table 11). At two ($p=0.03$) and six months of age ($p=0.05$) multiple regression (raw scores) showed that infants in the CMT group scored significantly lower on the AIMS compared with the control group. Infants who spent ≥three times daily in prone when awake had significantly higher scores on the AIMS than infants who spent less time in prone at two ($p=0.001$), six ($p <0.001$) and ten months of age ($p <0.001$) (Table 12 and 13). Gestational week, birth weight, birth length and plagiocephaly had no significant impact on the AIMS score.

Table 10. Alberta Infant Motor Scale (AIMS) percentile mean, SD, median and range at two, six and 10 months. P for group comparison.

Percentile at AIMS	CMT group	Control group
At 2 months of age		
Mean (SD)	20.4 (SD 18.4) P=0.02	31.8 (SD 21.5)
Median (Range)	12.0 (1 to 69)	23.0 (1 to 69)
At 6 months of age		
Mean (SD)	34.4 (SD 23.2) P=0.004	46.9 (SD 21.0)
Median (range)	27.0 (1 to 85)	41.0 (3 to 99)
At 10 months of age		
Mean (SD)	49.8 (SD 28.5) P=0.59	53.8 (SD 24.6)
Median (Range)	67.0 (1 to 100)	67.0 (1 to 100)

Table 11. Alberta infant motor scale (AIMS) scores ≤ 5:e and ≤10:e percentile at the ages of two, six and ten months.

Group and age (months)	≤5:e percentile	≤10:e percentile
CMT 2 m	10 % (n 3)	38 % (n 11)
Control 2m	3 % (n 1)	11 % (n 4)
CMT 6 m	9 % (n 6)	19 % (n 11)
Control 6 m	3 % (n 1)	3 % (n 1)
CMT 10 m	19 % (n 11)	23 % (n 13)
Control 10 m	3 % (n 1)	10 % (n 4)

Table 12. Result from multiple regression analysis (raw scores) at the ages of 2, 6 and 10 months.

Total number of infants in the analysis	Analysed infants	P values for group and for time spent in prone position at two months	P values for group and for time spent in prone position at six months	P values for group and for time spent in prone position at ten months
122	All infants independent of number of assessments	P=0.03 P=0.001	P=0.05 P=0.0000005	P=0.92 P=0.00000001
96	Those who missed one occasion.	P=0.01 P=0.003	P=0.06 P=0.000002	P=0.73 P=0.0000002
57	Those who participated on all occasions	P=0.02 P=0.004	P=0.04 P=0.007	P=0.52 P=0.03

First P value is for group and the second (below) P value is for prone time.

Table 13. Multiple regression, congenital muscular torticollis (CMT) and prone times awake effect on Alberta infant motor scale (AIMS) score. Number of times spent in prone when awake is dichotomized as ≤2 vs. ≥3. *Estimated difference in AIMS raw score between CMT and control, and for less and more numbers of times spent in prone awake.

	Standard errors	Beta*	P-value
At 2 months of age			
CMT	0.397	-0.252	0.03
Times spent in prone	0.495	0.389	0.001
At 6 months of age			
CMT	0.775	-0.177	0.05
Times spent in prone	0.848	0.472	0.0000005
At 10 months of age			
CMT	1.157	-0.009	0.92
Times spent in prone	2.254	0.521	0.00000001

Study V

The time needed to achieve satisfactory ROM in both rotation and lateral flexion was significantly (P<0.01 CI 1.4-2.8) shorter in the physiotherapist group than in the parents group (table 14). Symmetrical head position was achieved earlier (P=0.05) in the physiotherapist group with a median of 2.1 months and for the parent group the median was 4.1 months (table 14). The age at the start of treatment and start values for the affected side in rotation and lateral flexion had no significant influence on treatment time. There was an imbalance in muscle function between the affected and non-affected side for all infants at the start of treatment and for nineteen children there was still some degree of imbalance when full ROM and a symmetrical head position had been achieved.

Table 14. Treatment duration in median (range), mean and SD for physiotherapy and parent group.

	Physiotherapist group (months)	Parent group (months)
ROM in both rotation and lateral flexion	Median 0.7 (range 0.2 – 2.8) Mean 0.9 SD 0.7	Median 3.0 (range 1.0 – 9.0) Mean 3.1 SD 2.3
Symmetrical head position	Median 2.1 (0.2 – 5.3) Mean 2.5 SD 2.0	Median 4.1 (2.5 – 8.0) Mean 4.5 SD 1.9

DISCUSSION

General discussion

CMT can be estimated to occur in approximately 200 infants every year in the Gothenburg area and in approximately 2000 per year in Sweden. Stellwagen et al have reported that the incidence might be higher (2). CMT includes asymmetry in the early development of the head position, in ROM and muscle function of the neck and can affect craniofacial symmetry. The purpose of these studies was;

To undertake a survey of functional and cosmetic status in children with CMT.

To examine validity and reliability of the MFS.

To find reference values for rotation and lateral flexion of the neck and muscle function of the lateral flexors of the neck for a normally developing infant.

To investigate if infants with CMT are at higher risk of later achievement of early motor milestones compared to a control group of healthy infants.

To investigate if treatment duration is affected when stretching is done by an experienced physiotherapist compared with parents.

It seems that the prognosis for the condition is good. Most children treated for CMT in our study achieved an overall excellent/good status and the most notable findings were craniofacial asymmetry and asymmetry in muscle function i.e. strength/endurance. According to the parents problems in activities were rare. A delay in early motor milestones is rare after the age of 10 months. We also found that time spent in the prone position when awake seems to be of great importance in achieving early motor milestones.

Many studies show similar positive results in young children when treated for CMT before the age of one year (1,6,10,41,122). Whether their status will remain as good when they get older is hard to know as most studies are performed before the children have finished their skeletal length growth. In some follow-up studies on older children treated for CMT as infants the result is not as good (18,108), however most of these studies were performed several years ago. Both evaluation and treatment differ from the current study so we cannot generalize long-term result with reference to these “older” studies. It is known that untreated CMT can result in both functional and cosmetic problems (108,109,123,124,125) and having a mild CMT as an infant can give problems with discomfort and pain as an adult (108,124,125). In a study performed by Ippolitio and Tudisco three of the eight patients had pain in the affected

side radiating upward to their neck and face and downward to the shoulder and arm. The age of the patients at time of surgery was 24, 30 and 37 years (125). An extended follow-up period may be important (66,109) for children with remaining minor signs of CMT i.e. tendency of slight tilt of the head and/or a difference in muscle function of the lateral flexors between the right and left side. On the other hand we do not know how many infants that may spontaneously recover without any treatment (10). In clinic we have seen a high increase in infants referred for CMT during later year and this may indicate a higher prevalence of CMT or that health care professionals and/or parents are more observant. There is also a variation of number of births per year and the birth rate in our region has increased during the last years. However it can be speculated that there is a smaller number of infants with spontaneous recovery today than previously. As there seems to be a lack of time in prone position for many infants nowadays (84,85,86,87) this may indicate that milder cases of CMT resolved spontaneously in the past when strength in upper body and postural control was trained when more time were spent in the prone position.

ROM

We found that healthy infants had an excellent ROM of the neck in rotation and lateral flexion. Classification of PT can be a dilemma as the classification includes no limited ROM and there have been no available reference values for healthy infants. “Normal” ROM has been arbitrary which make the classification unsafe. ROM in rotation and in lateral flexion of the cervical spine has been measured by using several different methods (1,10,11,17,18,19,109,118,122). The method we used to measure rotation in sitting (18,19,109) was adequate for children in study I aged 2-5 years. When using this method it is of benefit if assistants can be available to hold the protractor. To measure rotation in infants in the supine position (1,41,103) is adequate but resource demanding as two people are needed to assist when measuring, one person to stabilize the infant’s shoulder and the other to hold the protractor. To achieve full ROM in rotation the examiner has to be able to rotate the infants’ head freely. If this is performed with the infants’ head resting on the surface of the examination couch the rotation will be limited. If measured with the head on the couch the chin will reach the surface and limit the rotation to approximately 70°-80° (118).

When measuring lateral flexion (18,19) it is of benefit to have the older children in supine as they relax more in lying than in sitting when measuring this movement. Additional staff is not needed when measuring lateral flexion in “older” children, as they mostly lay still and it is

easy for the evaluator to stabilize the shoulder. However, where there is a need for to the child to have assistance, one person is required to stabilize the infants' shoulders and it can be an advantage if an additional person distracts the infant during the measurements. If everyone involved in the measurements have the skills required the measurements do not take long and are easier to perform.

So-called normal values of a rather wide range have been stated in rotation 75-120° and in lateral flexion 40-90° (1,10,17,122,126). In physiotherapy practice it is common to consider 90° of rotation as normal/excellent PROM, but it is less than the reference value we found so we now consider 90° as slightly limited but not really abnormal. As a consequence of the reference values we found with a range of 100° to 120° (mean 110°), we can't really classify anyone with less than 100° of rotation as a "true" PT. The infants we classified earlier as PT often had a PROM in rotation of 90° in the affected side. However infants with only slightly limited PROM in rotation have shorter treatment duration than infants with more severe limitations and those with SMT (1,10,11). The question of classification needs further discussion and standardization of the inclusion criteria to assure that PT means the same in different studies.

This wide range of values considered as excellent or normal ROM for infants has a too large discrepancy to be acceptable either in a clinical situation or in research. Bartlett investigated ROM in the non-affected side in children with CMT (122) but no study on healthy infants was found in the literature. The term "normal values" should be abandoned as not everyone outside this range is abnormal (127), and people who have a particular condition may still fall within the "normal values". The term reference values are preferred as the reference population can be clearly defined. The differences in mean ROM in rotation that we found between healthy infants of two months of age and the ages of four, six and ten months are possibly due to the fact that it is not easy to gain cooperation from the very young infants when rotating their head. "Older" infants are more interested in the environment and are therefore easier to distract when performing the measurements.

There is a lack of reference values for neck PROM for pre-school children. It is possible that the decrease in ROM is greater during the early childhood years than the decrease in motion found by Youdas et al from the age of 11 years. Youdas et al investigated AROM in people

aged 11-79 years; they found that with increased age there was a change in both rotation and lateral flexion of the neck, in every 10-year period there would be an approximately lose of 3° (53). The reference value that Youdas et al found for AROM in the age group 11-19 years was; rotation mean 75° and lateral flexion mean 50° (53). Even when considering the fact that one expects AROM to be a little less than PROM there seems to be a lager decrease in ROM in early childhood i.e. before the age of 11 years.

Neck problems in adults are not uncommon (128,129,130) and it is unknown to what degree a remaining muscular imbalance from CMT may cause problems in adulthood.

Muscle function

The method used to measure muscle function in study I is found to be adequate for this age group and was used in two pervious studies of older children (18,19). Most of the children in study I were considered too young to be expected to cooperate with the use of a handheld dynamometer. In two studies with children above the age of six years a dynamometer was used to measure muscle function i.e. strength as a complement to the method used in study I (18,19).

In study III the MFS with five-degrees was already used to examine several of the infants before it was changed into the six-degree scale. We decided due to convenience to continue with "old" scale in that study. It is only the score 3 which is ambiguous, and could be either a 3 or a 4.

Most of the two months old infants were able to hold the head against gravity in a horizontal position and muscle function improved with age. Muscular imbalance between the right and left sides of the lateral flexors in the neck was very rare in healthy infants. In the reliability test of the MFS we used both weighted Kappa and ICC to calculate statistics, ICC is often used for reliability test (54,128,131) but there is a disagreement about the relevance to use it for inter-reliability. Even if both statistic methods gave similar results it would have been enough if we had only used the weighted Kappa. Also Bland Altman could have been used, the primary application of this statistic is to compare two clinical measurements but it can also be used to calculate inter and intra reliability. However weighted Kappa seems the best choice when it comes to the reliability of MFS as a discrepancy of one score is less problematic than the discrepancy of an increasing number of scores, the more discrepancy the less reliability.

We do understand that some practitioners will criticize the use of ICC and if we had only used ICC it would have weakened the study.

Muscle function is briefly described as part of the treatment in some studies (5,10,11,122), but it is rarely assessed (10) or evaluated (18,19). In a study on children aged 6-12 years who had been treated for CMT earlier there was a significant imbalance of muscle function between the affected and non-affected sides (18). More studies are needed to find out the importance of muscular imbalance/balance and also suitable treatment and duration of treatment. In connection with the validity discussion of the MFS it was changed to a six-degree scale. The scores 0,1 and 2 are the same, 4 has changed to a 5 and 3 can be either a 3 or a 4 when using the new scale. When using the six score scale it is only the "old score" 3 which is uncertain when comparing an infants muscle function with the reference values. Both the scores 3 and 4 are high over the horizontal line but at different levels. At two months of age most infants scored 1 on the MFS and this increased with age, there was no correlation with birth height or weight. Neither did Chiu et al find any correlation between muscle strength in the adult neck and weight or height (132). In healthy infants no difference between the two sides is to be expected in either ROM or muscle function of the neck.

Reference values for ROM in rotation and lateral flexion as well as reference values for muscle function of the lateral flexors of the neck provide more information for the physiotherapist. This can be of great value when evaluating, treatment and follow-up for infants with CMT. A reference value gives possibilities to clinically evaluate abnormal weakness or excessively power in the lateral flexors of the neck in infants. Hence the information and feedback to parents can improve. Standardized methods for measurements and estimates are necessary both in the clinical setting and in research and also to provide a possibility to compare the results from different studies.

Craniofacial asymmetry

Minor degrees of asymmetry of the face and the cranium are universal (4). More noticeable craniofacial asymmetry may occur during prolonged uncorrected head tilt (5,16,18,19,62,66,108). This is also one of the most notable remaining findings in follow up studies (11,18,19,66). The severity of craniofacial asymmetry is correlated to age and early treatment is important (16,71). It is probably individually how/if at all the asymmetries affects the quality of life. Some children/adolescents are clearly aware of the asymmetry.

Craniofacial asymmetries are seen more frequently now because more children are maintained supine for longer periods of time at early ages because the recommendations for the prevention of SIDS (57,61). The fear of SIDS also affects the awake position (86,133) and more has to be done to inform the parents of the benefits of prone time when the infant is awake (100,111,126).

Early motor milestones

Until the age of ten months the infants with CMT are of significantly higher risk of delay of achieving early motor milestones compared with the group of healthy infants, however time spent in a prone position when awake may have an impact on this.

The AIMS is reliable and valid in discriminating the motor performance of infants developing normally from infants' at-risk and motor delayed infants, and for evaluating small changes in motor skills due to maturation (89,90,134). However there seems to be a ceiling effect for AIMS once an infant can lower her- or himself in a controlled manner from a standing position (93). AS both our study and Schertz et al study (88) indicate that infants with CMT are at risk of having a delay in achieving early motor milestones it might be of extra importance for infants with CMT to spend time in prone position when awake to eliminate at least one risk factor. The prone position seems likely to be of particular importance in promoting the development for antigravity extension and gaining head control (133). It is generally accepted among physiotherapists and paediatricians that experience in a variety of positions are necessary for optimal motor development (80,81,82,83,84,85,86,87,133). We need to improve the information to make the parents realize the benefits of making the effort to give their infant more prone time. Today we often see in clinic that infants have somewhat low tolerance of prone position especially if it is not introduced early in life (84,86,87). It is possible that an infant with CMT has a lower tolerance than healthy infants due to the muscular imbalance in the neck. In our study also the control group was to some degree delayed in achieving early motor milestones at two months of age and plagiocephaly was also more common than expected. Maybe this is a result of a lack in sufficient information about prone time awake after the back to sleep campaign (84,85,86,133). Parents of a newborn have a lot of important issues to deal with i.e. feeding, sleeping etc. so it is understandable that information about time in prone may be neglected. The fear of SIDS also affects the parents' choice of position for their infant when awake (86,133).

It is observed that an infant with CMT can have a more limited repertoire than a healthy infant during the first months of life (11). The CMT group had significantly lower scores at AIMS than the control group in the current study but the fact that both groups were below the 50th percentile at AIMS at two months of age may have camouflaged some possible differences in repertoire between the groups. One thought is that there could have been benefits to first recommend prone time awake in order to promote motor development and then perform the assessment at a slightly older age for example at three months of age.

Stretching treatment

In the randomize treatment study there where more clinical appointments for one of the groups and it was hard to totally blind the evaluator from which group the infants belonged to. As the evaluator occasionally met some of the infants in the waiting room and if she saw a particular child often it would be obvious that this infant probably belonged to the physiotherapy group. The optimum circumstances would have been if the evaluator never had a chance to see the infants between the evaluation appointments but this was not practicable in reality. However the evaluator was independent and although she may have been able to guess which group the infants belonged to we have no reason to believe that this influenced the measurements. Infants with CMT gained full ROM and symmetrical head posture earlier when treated by an experienced physiotherapist compared to parents. Nevertheless parents can achieve a good result within a couple of months.

It may be harder for parents to accomplish the stretching treatment, as the study showed that parents needed more than twice the time of an experienced physiotherapist to gain full ROM. In most studies where the parents perform the treatment and the mean duration time is about 4.7 months (10). The parent group gained a rather short treatment duration compared with other studies on home treatment. This study is small but it indicates that parents can perform the stretching exercises with a good result and treatment time can be shorter than previously reported. The challenge for the physiotherapist is to find and give the “best” instructions, encouragement and support based on the prerequisites of the individual child and family to help the parents to perform as optimally as possible.

CONCLUSIONS

- Most children who were treated for CMT earlier in this study had an overall excellent/good status and the most notable findings were craniofacial asymmetry and asymmetry in muscle endurance. Problems in activities were rare.

- The MFS was found to have construct and content validity to measure muscle function of the lateral flexors of the neck in infants with CMT. The inter-rater and intra-rater reliability is high for both novice and experienced physiotherapists when examinening photos.

- Reference values for PROM in infants aged 2-10 months neck ranged from 100-120° (mean 110°) for neck rotation and from 65-75° (mean 70°) in lateral flexion. Muscle function in lateral head righting as measured by the MFS demonstrated that most two-month-old infants, when held in a horizontal position, were able to maintain the head in line with their bodies. By ten months-of-age, the infants were able to hold their heads either high or very high above the horizontal line.

- The CMT group were of significant risk of delay in achieving early motor milestones compared with the control group until the age of ten months, but time spent in a prone position when awake seemed to be of greater magnitude than the CMT. Gestational week, gender, birth weight, birth length and plagiocephaly had no influence on motor development in this study.

- Infants with CMT gained full ROM and symmetrical head posture earlier when treated by an experienced physiotherapist compared to parents. Nevertheless parents can achieve a good result within a couple of months.

CLINICAL IMPLICATIONS

The MFS has made it easier to assess and evaluate muscle function in infants with torticollis. It is also a tool that can be used to give parents feed back after training of muscle function i.e. if there is progress or not. The publishing of the reference values made the MFS available to more physiotherapists than previously. The reference values of ROM and muscle function give an indication of what to expect of the infant at a certain age.

Infants in general and infants with CMT in particular needs a lot of time in the prone position when awake to stimulate motor development, postural control and decrease the risk of developing plagiocephaly. To promote better information about prone time (and plagiocephaly) we have provided education for the staff at the child health centre in our area. A pamphlet with information about prone time has been produced for parents with a newborn and is distributed via the child health centre. This pamphlet is also circulated via physiotherapists working with infants through a network consisting of Swedish therapists interested in CMT. A website for torticollis is also used to spread information but more ways to disseminate this information is needed in order to reach as many parents of newborns as possible.

There is an indication for a physiotherapist to perform the stretching treatment in difficult cases i.e. great limitation of ROM and/or a resistant infant. If the physiotherapist can achieve a satisfying ROM in rotation of the neck of the infant earlier this might prevent that the infant from developing a severe plagiocephaly.

FURTHER STUDIES

These studies give some answers but they also raise new questions and it would be desirable to do a follow up study on infants with CMT at an older age when the skeletal growth is finished. The effect on the cervical spine long term from remaining head tilt and/or muscular imbalance needs to be investigated. The effect of different treatment approaches on muscle function is currently part of an ongoing study. A follow-up study of motor development at preschool age is planned to start during 2009. It would also be very interesting to perform research involving evaluation of muscular tissue and function at different stages of CMT.

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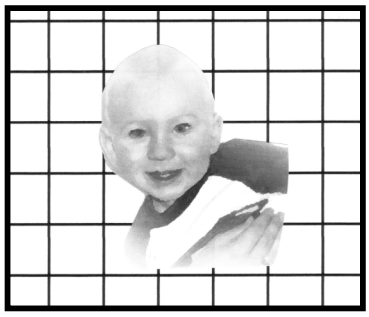
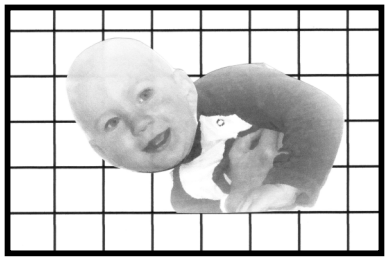
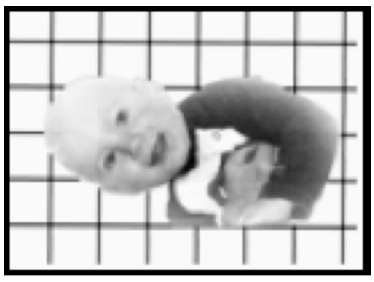
APPENDIX A



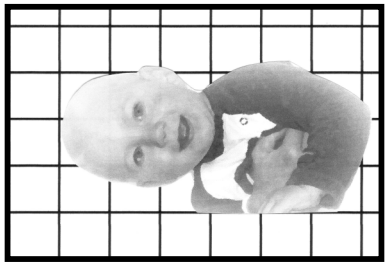
4. Very high over the horizontal



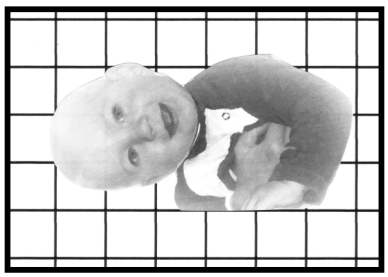
3. High over the horizontal



2. Briefly over the horizontal



1. In the horizontal

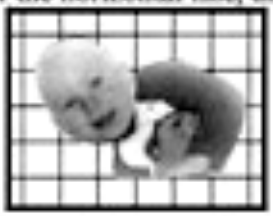


0. Below the horizontal

APPENDIX B

Muscle function scale (MFS) for infants

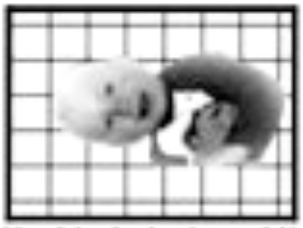
5. Head very high over the horizontal line, almost vertical position.



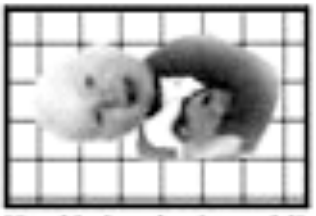
4. Head high over horizontal line and more than 45°.

3. Head high over horizontal line but below 45°.

2. Head slightly over the horizontal line.



1. Head in the horizontal line.



0. Head below horizontal line.