

RESEARCH REPORT

Evaluation of treatment strategies for muscle function in infants with congenital muscular torticollis

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ABSTRACT

This study focuses on the treatment of the muscle function imbalance and asymmetric head posture for infants with congenital muscular torticollis (CMT). The aim of this study was to compare treatment time for groups with different strategies for muscle function training. The treatment goal was to achieve a symmetric head posture. Thirty-seven infants were randomised to three groups. Group I was treated only with handling strategies. Group II got the same handling strategies but also received specific strength exercises. Group III received the same treatment as group II but was also provided with weekly training by a physiotherapist. Mean age at the start of treatment was 4.5 months of age, range 1–10.5. The possible effect of covariates was also investigated; age at the start of treatment, range of motion (ROM) in rotation of the neck, ROM in lateral flexion of the neck, the muscle function scale (MFS) score, plagiocephaly, and gender were analysed with ANCOVA. Thirty-one treated infants achieved symmetric head posture before the age of 12 months. Mean treatment time (3.5 month) did not differ significantly between the groups. All infants randomised to group I could stay in that group. The MFS score and age at the start of the treatment influenced treatment time ($p < 0.05$). The treatment time for all groups was similar. Early referral to physical therapy of infants with CMT and muscle function imbalance in lateral righting could shorten treatment time. Studies about natural course and long-term effects of muscular imbalance must be investigated in future research.

INTRODUCTION

Congenital muscular torticollis (CMT) is the third most common musculoskeletal abnormality in infants next to hip dysplasia and clubfoot. The reported incidence is 0.4–2.0% (Cheng et al, 2000); however, a recent study indicates that it might be higher (Stellwagen, Hubbard, Chambers, and Jones, 2008). CMT is a result of shortening or excessive contraction of the sternocleidomastoid (SCM) muscle with limited range of motion (ROM) in both rotation and lateral flexion in the neck. The head is typically tilted in lateral flexion toward the affected SCM in

the frontal plane and rotated toward the opposite side in the transversal plane (Cheng et al, 2000; Ho, Lee, and Singh, 1999). Because of the positional preference, there is a high risk that infants with CMT develop deformational plagiocephaly (Golden, Beals, Littlefield, and Pomatto, 1999; Hummel and Fortado, 2005; Oh, Hoy, and Rogers, 2009; Rogers, Oh, and Mulliken, 2009). During the time of skeletal growth, the remaining CMT can have an effect on facial growth, with the result of more or less obvious facial asymmetry (Itio et al, 1990; Minamitani, Inoue, and Okuno, 1990; Stassen and Kerwala, 2000). The head tilt itself is assumed to cause the facial asymmetry (Greenberg and Pollard, 2000). There is an imbalance in muscle function around the neck for infants with CMT; it has been found that the lateral head righting on the contralateral side is weakened compared to the affected side (Binder, Eng, Gasier, and Koch, 1987; Cheng and Au, 1994;

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Emery 1994). Our clinical experience is that the affected side seems to be excessively stronger and the contralateral side weaker than expected for the age. This imbalance in the neck muscles has not been found in healthy control subjects (Öhman and Beckung, 2008).

General treatment goals are to resolve the limitations of neck movements and muscular imbalance; prevent facial and skull deformities; and prevent long-term posture changes (Do, 2006; Emery, 1994; Minamitani, Inoue, and Okuno, 1990). The treatment, assessment, and evaluation of muscle function imbalance in infants with CMT are only described briefly (Emery, 1994). Muscular imbalance has been seen in older children who had CMT as infants (Öhman and Beckung, 2004; Öhman and Beckung, 2005). Our clinical experience is that it can be hard for a child to achieve symmetric head posture when there is a remaining imbalance in muscle function. It can be of great importance to reduce muscular imbalance in young infants with CMT. Generally, both handling strategies and specific strength exercises have been given for home treatment for infants at our clinic. The head righting reaction is used to stimulate muscle function in both handling and exercises. Some parents seem to focus mostly on handling strategies, and some parents forget this and focus more or less on the specific strength exercises. However, we have noticed that with parents who have only used handling strategies consistently and intensively that the effect has been very good. This has raised the question if it might be better to focus more on the handling strategies at the start of treatment for infants. This study focuses on the treatment of the muscle function imbalance and asymmetric head posture for infants with congenital muscular torticollis (CMT).

The aim of this study was to compare treatment time for groups with different strategies for muscle function training and to investigate if it is defensible to use handling strategies as the primary treatment. The treatment goal was to achieve a symmetric head posture. The covariates investigated were age at the start of treatment; ROM in rotation of the neck; ROM in lateral flexion of the neck; the muscle function scale (MFS) score; plagiocephaly; and gender.

METHOD

Study design

Inclusion criteria were that the infants were diagnosed by a paediatrician to have CMT and were referred for physiotherapy. Those infants who had other known

conditions or diseases were excluded. The families were asked to participate in the study before commencing treatment. The families who agreed to participate in the study were randomised to one of three groups; directly after they had agreed to participate the parents were asked to pull a sealed envelope from a stack, which identified their group. Group I got handling strategies only, and groups II and III received both handling strategies and specific strength exercises. The reason for including two groups with the same exercises (group III was offered extra help with the training by a physiotherapist two or three times a week) was to ensure that at least one group did both the handling strategies and the specific strength exercises. Assessment of ROM, muscle function, head position, and plagiocephaly were performed once every month and always by the same physiotherapist; the evaluator was not blinded. Two other physiotherapists gave instructions to all groups and provided help with treatment in group III. All parents were given written instructions.

Participants

During a period of about 6 months 42 families were asked to participate in the study. Five families chose not to participate in the study due to the inconvenience if they needed to travel to the hospital often. The parents of 20 girls and 17 boys agreed to participate. The infants were randomly assigned to group I, II, or III. There were four dropouts. The parents of one infant changed their mind after being randomised to group III, and one stopped because of family reasons. Two infants were excluded after starting the treatment because it was discovered that they had mild paroxysmal torticollis.

Procedure

The importance of consistency in handling strategies was explained to all groups. All parents were asked to make notes of exercises they performed in a handling/training diary. All parents were also allowed additional appointments with the physiotherapist if they felt unsure of how to perform the treatment. If any of the infants in group I had an obvious lack of improvement after the first 2 months, it was decided that they would be given the same treatment as group II. This is the treatment generally given at the clinic. The reason for this was to ensure that all infants had an efficacious treatment in the long-term. Stretching exercises were included in the treatment for infants with limited ROM (<90° in rotation and/or side difference in lateral flexion). Information

on how to prevent and reduce plagiocephaly was given to all participating parents.

ROM was measured in the degrees of rotation with an arthrodial protractor (Cheng, Tang, and Chen, 1999; Cheng et al, 2000; Öhman and Beckung, 2008), with a reference value of 110 degrees (Öhman and Beckung, 2008). The degree of lateral flexion was measured with the infant lying supine on a large protractor (Öhman and Beckung, 2005; Öhman and Beckung, 2008; Perbeck-Klackenberg, Elfving, Haglund, and Brogren-Carlberg, 2005). The reference value for lateral flexion is 70° (Öhman and Beckung, 2008). Muscle function was estimated with the 6° MFS, which has an intra/interreliability Kappa > 0.9 (Öhman and Beckung, 2008; Öhman, Nilsson, and Beckung, 2009). The scores for both sides were noted. Head tilt (neck involvement) and plagiocephaly (posterior flattening and/or forehead asymmetry) were assessed by using three of the five items of the “Severity Assessment for Plagiocephaly.” Each of these three items (neck involvement, posterior flattening, and forehead asymmetry) is scored on a four-point scale of 0–3 (neck involvement 0 = no head tilt-symmetric head posture; 1 = minor head tilt; 2 = moderate head tilt; and 3 = severe head tilt). Each item was analysed separately (Losee and Mason, 2005). The local ethical committee in Gothenburg approved the study, and the parents gave their informed consent.

Intervention

Handling strategies

The infant had to spend as much time as possible in a prone position when awake and supervised (Figure 1). A lot of strategies for compliance to prone position in different ages were given to all parents both by the physical therapists and through written information (a brochure we made earlier for the child health care centers in cooperation with paediatricians). The infant had to be held in a sideways position with the weaker side facing upward (the “non-affected” side). This was the case whenever lifting the infant up or



FIGURE 1 Prone positioning while awake.



FIGURE 2 Handling position when lifting, focusing on holding the infant in a sideways position with the weaker side facing upward.



FIGURE 3 Handling position during holding and carrying, focusing on a tilted (45°) position with the weaker side facing upward.

putting him/her down (Figure 2). If possible, this procedure was performed slowly, and only if the infant was able to hold its head up. Whenever an infant was carried, it had to be held in a tilted position (approximately 45°) with the weaker side facing upward (Figure 3). When suitable, rotation of the head toward the affected side should be stimulated. This should happen often.

Specific strength exercises

While sitting on the parent’s knee, the infant was tilted to an almost horizontal position with the weaker side facing upward and held for 5–15 seconds. This was repeated over a 15-minute period with breaks when needed (Figure 4). With the weaker side facing upward and when standing in front of a mirror, the infant is lowered to a horizontal position and held for 5–10 seconds. This was repeated over a 15-minute period with breaks when needed (Figure 5).

Data analysis

There were descriptive statistics for the age at the start of treatment, treatment time, plagiocephaly,



FIGURE 4 Strengthening exercise during sitting, focusing on tilting the infant to an almost horizontal position with the weaker side facing upward. The maneuver should be held for 5–15 seconds, repeated with rest breaks over a 15-minute session.



FIGURE 5 Strengthening exercise during standing, focusing on tilting the infant to an almost horizontal position with the weaker side facing upward. The maneuver should be held for 5–10 seconds, repeated with rest breaks over a 15-minute session.

and gender. Differences in treatment time between groups were analysed with multiple regression analyses using an ANCOVA model with the covariates of age at the start of treatment; ROM at start of

treatment; MFS score for both sides at the second assessment; plagiocephaly score from “Severity Assessment for Plagiocephaly”; and gender.

RESULTS

Thirty-three infants participated: 9 in group I; 13 in group II; and 11 in group III. The mean age at the start of treatment was 4.5 months of age, range 1–10.5. All infants had a difference in MFS score between affected and not affected side. The affected side had the higher score. The mean difference between affected and not affected side was 2 scores, range 1–3 (Table 1). The mean treatment time was 3.5 months, range 1–5.5. There were no significant differences between the treatment groups in the time needed to achieve a symmetrical head posture (Table 2). No infant in group I had to change his or her treatment group because they all achieved symmetrical head posture. With this result we decided to stop including any more infants in the study. Multiple regression analyses using an ANCOVA model showed that the MFS scores at both sides ($p < 0.01$ and $p = 0.03$) and the age at start of treatment ($p = 0.04$) had significant influence on treatment time. The covariates, limited ROM in lateral flexion and rotation, plagiocephaly, and gender, had no significant effect on treatment time. For the 31 infants who attained symmetric head posture during the study, 19 (61%) achieved symmetrical scores on MFS. Eleven (36%) still had a difference of one score, and one infant had a difference of two scores. Two infants had a remaining limited ROM in rotation when they had achieved a symmetric head posture. They continued with stretching treatment and follow-ups. Four

TABLE 1 Data at start and at the end of the study for all of the 33 infants who participated.

	At start of study	At the end of the study All 33 infants (included the 2 infants who had a break in treatment)	At the end of the study The 31 infants who achieved symmetric head posture during the study
Head tilt	33 (100 %)	2 (6 %)	0
Difference in MFS scores	33 (100 %)	12 (36 %)	10 (32 %)
Limited ROM <90° in rotation	10 (30 %)	2 (6%)	2 (6%)
Limited ROM in lateral flexion	7 (21 %)	0	0
Plagiocephaly	22 (67%)	2 (6 %)	1 (3 %)
Age at start of treatment	4.5 months (range 1–10.5)		
Treatment time			3,5 months (range 1–5.5)
Gender	16 girls 17 boys	16 girls 17 boys	15 girls 16 boys

TABLE 2 Multiple regression, effects of group, and the covariates age at the start of treatment, ROM in rotation of the neck, ROM in lateral flexion of the neck, and the MFS score.

	Standard errors	B	95 % confidence interval	p-value
Age at start of treatment	0.158	-0.404	-0.671 to -0.016	0.041
Group	0.488	-0.632	-1.749 to 0.274	0.145
ROM in lateral flexion at start of treatment	0.060	0.328	-0.055 to 0.194	0.261
ROM in rotation at start of treatment	0.026	-0.036	-0.09 to 0.018	0.178
MFS score in affected side	0.475	0.790	0.712 to 2.682	0.002
MFS score in contralateral side	0.408	-0.434	-1.830 to -0.137	0.025

β is the standardised regression coefficient.

infants still had minor plagiocephaly when a symmetric head posture was attained. The quality of the notes in the handling/training diary differed. Some parents had made very specific notes while some made more general notes. Others only made notes when they had missed the treatment. Seven families, according to their notes, trained less than expected (i.e., less than 5–7 days a week) all belonging to groups II or III.

Two infants, one in group II and one in group III, still had a tendency to tilt the head when the physiotherapist decided to take a break in treatment. This break was because the infants were close to 1 year of age, an age when it can be hard to get infants to cooperate. For these two infants the treatment time could not be analysed.

DISCUSSION

In the current study no infant in group I had to change group because all infants in this group achieved symmetric head posture. This indicates that our theory that the treatment strategy with consistent handling for infants with CMT can work well as primary choice of treatment for muscle function. The contralateral weaker side sometimes is very weak at first assessment, and the MFS score does not differentiate the weakness below score 1. That's why we chose to use the MFS score from the second assessment in the analysis to avoid a type II error. We believe that for some parents too many exercises can be overwhelming. In the clinic there are parents who acknowledge that they only partly perform the treatments they are supposed to give their infant. Our impression was that some parents in group III did less at home because they felt confident that they could do less as they got help with the training. This is partly confirmed as seven families according to the training diary did less than expected, all from groups II and III. If most of the training can be integrated into daily routines, it may prove easier for the parents to

accomplish optimal treatment. Consistent training integrated into daily routines will probably give a lot of muscle function training during the day. This might produce more strength exercise for the neck than specific exercises performed at one or two times daily. If the hypothesis, "that it is easier to be consistent with a smaller number of different handling strategies/exercises to perform" is true, this might explain why there were no significant differences between the groups.

Limitations

The handling strategies/training diary did not work optimally, and not all parents followed the instructions on how to document. It can be argued that it may have been a better idea to ask the parents how the treatment worked at home. This could have been done either by phone or during their visits to the clinic. Close email contact with the parents might be an efficient way to both get information and stimulate compliance with the training program.

Another limitation is that we had no control group without any treatment. We do not believe that any of the parents would have accepted their infant being in a group without any treatment. We could have chosen to have a group without treatment for some months, and if there was no improvement, we could have then given them treatment later; but with older infants it is often harder to get cooperation, and this may have extended the treatment time considerably. In our experience, with infants who come at a later age (i.e., after 8–9 months of age), it is not uncommon that breaks in treatment are required, with a new treatment starting at around 2 years of age, when the child can often show a much better level of cooperation, allowing for new and more fun exercises to be introduced. However, if we had chosen a group with delayed treatment, this might have given us valuable additional information. This will be considered in further studies. Shorter time between the assessments

and a blinded evaluator also would have strengthened the study.

The covariates of MFS score and age at the start of treatment had a significant influence on the treatment time. This indicates prognostic information about treatment time required to achieve symmetrical head posture. It has been found in other studies that the age at the start of treatment of CMT affects the result (Cheng et al, 2000; Cheng et al, 2001). It could be of great importance to encourage early referral to physical therapy for infants with CMT. Gains for both the individual infant and the health care system are possible. Not all infants in our study reached equal scores on the MFS in the time needed to achieve a symmetric head posture. Healthy infants have been found to have a balance between MFS scores in left- and right-hand sides (Öhman and Beckung, 2008). In most infants in our study who had unequal MFS scores at the end of the study, the difference was only one point. If the infants accept it, the handling strategies should be continued after symmetric head posture is obtained. Muscle function balance could be obtained at a later stage. Further studies are needed on how muscular imbalance might affect the neck and head position in the long term.

Clinical implications

The new guidelines contain a primary program with “treatment in handling” (Appendix 1) and a secondary program with “specific strength exercises” (Appendix 2). We expect that most infants will probably only need the primary program. This is if the parents are consistent in their handling strategy. To ensure that the new guidelines give expected results, we plan to perform a retrospective study in about 2 years. This will be after the new guidelines are fully integrated in the clinic. It is our belief that treatment in handling will act as an improved prerequisite for compliance because it is perhaps easier for parents to “understand” and easier for them to integrate into their daily lives.

CONCLUSION

The treatment time did not differ significantly between the treatment groups. The MFS score and age at the start of the treatment had a significant influence on time to achieve symmetric head posture. Early referral to physical therapy of infants with CMT and muscle function imbalance in lateral righting can shorten treatment time. Studies about natural course and long-term effects of muscular imbalance must be investigated in future research.

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Appendix 1. Protocol for training of muscle function in daily activity for infants with left-sided torticollis (treatment handling).

The muscle with weaker activity that needs training is the big neck muscle on the right side, which side bends the head to the right and rotates it to the left. Be consequente in stimulating the musculature at the right side; use any opportunity in daily life. Doing this will give most infants a good chance to achieve a good muscular function.

To give the infant a lot of tummy time when awake is beneficial for head control and neck musculature. Please make it a habit to always put your child in prone when awake even if it is only for a short moment.



Stimulate your child to rotate the head to the left side because it gives training to the big neck muscle that is weaker.



When you stand or sit with your child in your arms, you can use the child's curiosity and put yourself in a position that make the child to look at her/his left side.



Every time you lift your child up from lying, do this in the side position. Take your child up with the left side downward. It is best if the baby looks down (straight forward is ok but not upward)! Use the same method when you put your child down. To raise the challenge the exercise can be done slowly.

When you carry your child, hold it a little tilted with the left side downward, even if it is only for a short moment.



Appendix 2. Training of muscle function for infants with left-sided torticollis (specific strength exercises).

The large neck muscle on the right side is the weaker muscle that needs training. This bends the head to the right side and rotates it to the left.



With your child positioned on your knee, tilt her/him to the side to an almost horizontal position, with the weaker (right) side up. Hold for 5–15 seconds. Repeat for about 15 minutes, taking breaks when needed.



Lower your child to a horizontal position with the weaker (right) side up, without any support for the head. Stand in front of a mirror. It is better if your child also looks down (rotates the head to the left). Hold for 5–10 seconds followed by a rest of 5 seconds. Repeat for about 10–15 minutes, taking breaks when needed.

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