Counter -positioning +/- physiotherapy or helmet therapy may reduce skull asymmetry in the infant with plagiocephaly. The most effective conservative interventions for torticollis and plagiocephaly cannot be determined.

Reviewer: Peggy Goff PT (pgoff@thc.on.ca)
Date: October 2006

Clinical Scenario
Infants with torticollis +/- plagiocephaly are commonly seen within a physiotherapy department for assessment, ongoing follow-up and for parental instruction in a home program consisting of gentle passive stretching, stimulation of active ROM, positioning to minimize skull and facial asymmetry and facilitate motor development. The incidence of plagiocephaly in infancy has increased since the introduction of the Back to Sleep campaign to reduce the incidence of SIDS.

Focused clinical question
What are the most effective and efficient non-surgical treatment interventions (with respect to content, frequency, duration and considering parental and infant distress) for the infant with Congenital Muscular Torticollis with and without plagiocephaly that will maximize treatment outcomes (head tilt, ROM, head shape, motor development)?

Summary of Best Evidence
No randomized clinical trials were found. One systematic review was found.
For the purposes of this CAT the following is the summary of best evidence.
• Cheng et al, 2001 in their large prospective study with limitations in its methodology, showed that controlled manual stretching by a therapist and positioning/stimulation is safe and effective in the treatment of congenital muscular torticollis in about 95% of patients who are first seen before the age of one year. The most important factors to predicting outcome are clinical group, initial deficit of rotation, and age at presentation.
• Bialocerkowski et al, 2005 in their systematic review on conservative interventions for the treatment of plagiocephaly concluded that it is not possible to recommend the most effective conservative interventions due to poor study methodological quality and bias. There is no standardized outcome measure to quantify skull shape or magnitude of asymmetry. Counter positioning +/- physiotherapy or helmet therapy may reduce skull asymmetry. Further investigation is warranted but should be preceded by the standardization of outcome measurement tools.

Clinical Bottom Line
It is not possible to determine the content or frequency of the most effective treatment strategy for torticollis/plagiocephaly due to poor study methodology and lack of valid and reliable outcome measures. With the lack of randomized controlled trials it is uncertain what role time and the natural progression of the condition plays in the outcome. Previous studies have concluded that parental stretching is effective but have also lacked methodological quality.
To allow a more consistent assessment and patient evaluation between therapists and to perhaps aid in prediction of duration and outcome I would recommend
• Therapists classify the torticollis patient into the 3 classifications (SMT, MT, PT)
• The inclusion of rotation and side flexion limitation, head tilt, residual band, asymmetry, parental subjective cosmetic/functional status and the measurement of muscle endurance to ensure a standardized and comprehensive assessment
• Clinical category, age at presentation and limitation of rotation at presentation are potential triggers for the possibility of a poor outcome so those clients may need to be evaluated more often to ensure parental treatment is effective.
• Therapists should be made aware that there have been reported cases of the sternomastoid snapping during stretching with no long-term implications.
Limitation of this CAT- This critically appraised topic has not been peer-reviewed.

SEARCH STRATEGY:

Terms used to guide Search Strategy:

- **Patient/Client Group:** Participants- infants with congenital muscular torticollis with or without plagiocephaly
- **Intervention (or Assessment):** range of motion exercises, manual therapy, stretching, positioning, stimulation, helmet therapy
- **Comparison:** torticollis with or without plagiocephaly
- **Outcome(s):** ROM, head posture, head shape, motor development

<table>
<thead>
<tr>
<th>Databases and sites searched</th>
<th>Search Terms</th>
<th>Limits used</th>
</tr>
</thead>
</table>
| Medline, CINAHL, Healthstar, EBM reviews, Cochrane Library, PEDro, National Guidelines clearinghouse, Uptodate | **Subject headings**
Torticollis, Infants, Therapeutics, Treatment Outcomes, Orthoses, Craniofacial dysostosis,

**Key words**- congential muscular torticollis, plagiocephaly, stretching, helmet therapy | **Inclusion Criteria**
- meta analyses, systematic reviews, randomized controlled trials, cohort studies, case control and case series
- English
- 2000-2006
- Humans |
RESULTS OF SEARCH

Table 1: Summary of Study Designs of Articles retrieved

<table>
<thead>
<tr>
<th>Study Design/ Methodology of Articles Retrieved</th>
<th>Level</th>
<th>Number Located</th>
<th>Author (Year)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cross-sectional</td>
<td>3</td>
<td>1</td>
<td>Ohman, and Beckung, (2005)</td>
</tr>
<tr>
<td>Systematic review (Not of RCT)</td>
<td>3</td>
<td>1</td>
<td>Bialocerkowski, et al (2005)</td>
</tr>
<tr>
<td>Narrative review</td>
<td>4</td>
<td>2</td>
<td>Van Vlimmeren, et al (2006), and UpTo Date</td>
</tr>
</tbody>
</table>

BEST EVIDENCE

The following 2 papers were identified as the 'best' evidence and selected for critical appraisal. Reasons for selecting these studies were:

- No randomized controlled trials were found.
- The Bialocerkowski et al 2005 article was included as it was a systematic review of the plagiocephaly topic and provided a critical analysis of many studies.
- The Cheng et al 2001 article was chosen as it was the prospective study with the largest sample and provided some insight into the predictors of treatment outcome.

SUMMARY OF BEST EVIDENCE


Aim of Study
To evaluate the effectiveness of a standardized manual stretching program on a large group of patients with clearly defined congenital muscular torticollis and to evaluate factors that predict outcome of treatment
1. Population:
1086 patients with congenital muscular torticollis ages 0-1 year who had been seen at the Torticollis clinic from 1985 –1997
• 821 included in the study
• 237 were excluded 117 >1 year old and 120 had another type of torticollis
Patients were classified into
• 3 clinical diagnoses sternomastoid tumor (SMT) muscular torticollis (MT) and Postural torticollis
• Those with ROM rotation limitation <= 15 degrees and >15 degrees
• Ages- < 1 month, 1-3 months, 3-6 months, 6-12 months

2. Intervention
For those who presented with a >10 degree passive rotation limitation
• Manual stretching program performed in the clinic by a physiotherapist only
• 3 times per week, 3 repetitions times 15 stretches of the tight muscle 1 second hold, 10 second rest
• Home program of stimulation of active ROM and positioning
For those with < 10 degree limitation
• Home program of stimulation of Active ROM and positioning only

3. Outcomes
Outcomes measures included
• Total duration of treatment
• Overall score of measurements that included
  o Rotation deficit, lateral bending deficit, residual band (none, lateral, cleidal, sternal), head tilt, subjective assessment (cosmetic and functional) by parents
• Need for operative treatment

4. Evaluation
• 821 were treated in study
• 788 (96%) were available for overall final scoring
• Evaluation was at initial assessment and at final follow up in clinic
• Mean duration of follow up was 4.5 years (1.5-13 years)

Results
Clinical Classification-
• Sternomastoid tumour SMT 55%, Muscular torticollis MT 34%, Postural torticollis PT 11%
• Right side affected 49%, left side affected 51%
• Male : female 3:2 in all groups

Age at presentation
• patients in SMT group presented earlier than other groups p,.0001

Limitation of ROM-
• rot>15 deg more prevalent in SMT group p,.0001
Higher rate of breech and vacuum extraction <.0001
Greater rate of hip dysplasia <.0002
Giving way or snapping of SMT occurred in 8% of patients and the risk was greater if
• hip dysplasia, left side affected, rotation >15 degrees and < 1 month at presentation

Duration of treatment
• Significant difference between SMT/MT and SMT/PT groups <. 0001
  • SMT 3.7 months- median
• MT 2.5 months- median
• PT 1.4 months- median

Duration of treatment was associated with
• Rotation deficit > 15 degrees < .0001
• Clinical group p< .0001
• Age > 1 month at presentation p<.0001
• Right side affected p< .0001
• Difficult birth p< .0001

Results at final evaluation

<table>
<thead>
<tr>
<th>Overall score</th>
<th>SMT</th>
<th>MT</th>
<th>PT</th>
<th>groups</th>
</tr>
</thead>
<tbody>
<tr>
<td>Excellent/Good</td>
<td>87.8 %</td>
<td>93.8%</td>
<td>98.9 %</td>
<td></td>
</tr>
<tr>
<td>Poor/Fair</td>
<td>12.2 %</td>
<td>6 .2%</td>
<td>1.1 %</td>
<td></td>
</tr>
</tbody>
</table>

The overall score was associated with
• rotation deficit >15 p<.02, age 1-3 months at presentation p<.014, duration of treatment p<.0001

Rate of surgical intervention
• SMT 8% had surgery, MT group 3 % had surgery, PT 0% (not significant difference between PT and MT)

Significant risk factors for surgery-
• late age at presentation, clinical group SMT and rotation deficit >15 degrees

Critical Analysis

Study Design- Prospective longitudinal case series
• no justification of sample size or informed consent
• no comparison group/control

Reliability and validity of outcome measures-
• There was no documentation as to the validity of the rating scale used for the overall score. The only notation as to reliability was for the measurement of rotation by an arthrodial . The rating scale was cited as used in many other studies but it is not known if its validity has been tested.

Intervention- intervention was not documented clearly enough to allow repetition of the treatment. The follow up averaged 4.5 years but range was large 1.5-13 years
Only 96% were available for final follow-up and no discussions as to reasons for dropout

Results- most results reported were highly significant however were based on outcome measures that may not be valid

Conclusions- concluded that controlled manual stretching by a therapist and positioning/stimulation is safe and effective in the treatment of congenital muscular torticollis in about 95% of patients who are first seen before the age of one year. The most important factors to predicting outcome are clinical group, initial deficit of rotation, and age at presentation. The need for surgery is associated with late age at presentation, clinical group SMT and rotation deficit >15 degrees.

Citation
Summary/ Critical Analysis

• A systematic review on the effectiveness of conservative interventions for the infant with plagiocephaly was performed
• No randomized controlled trials were found and 16 studies (comparative, prospective and retrospective case series) were selected for review
• Articles were appraised using the Law, 2002 Critical review form - quantitative studies and most were found to be moderate to poor in quality
• Data was provided in a narrative format and a meta analysis was not provided due to types of studies found
• No conclusions as to the effectiveness of the conservative interventions could be made due to lack of both methodological quality and valid/reliable outcome measures. There is a need for the development of valid and reliable outcome measures prior to further research in this area
• Counterpositioning +/- physiotherapy or helmet therapy may reduce skull asymmetry

Implications for Practice

Therapists are unable to select specific treatment interventions based on evidence, as effectiveness has not been determined. The knowledge of the possible predictors of outcome may be useful in planning the treatment program and frequency of follow up. Parental home programs of manual stretching are commonly used in practice for this population. Prior low quality studies have shown this type of intervention to also be effective. Due to the lack of comparison studies we are unable to determine if parental or therapist stretching are equally effective. Referral for assessment for helmet therapy or surgical intervention should be made for those not responding to the treatment program with ongoing limitations of range of motion and asymmetry.

References
