Clinical Determinants of the Outcome of Manual Stretching in the Treatment of Congenital Muscular Torticollis in Infants

A Prospective Study of Eight Hundred and Twenty-one Cases

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Background: The natural history of congenital muscular torticollis and the outcome of different treatment modalities have been poorly investigated, and the results of treatment have varied considerably.

Methods: The main objective of this prospective study was to evaluate the outcomes of 821 consecutive patients with congenital muscular torticollis who were first seen when they were less than one year old, were treated with a standardized program of manual stretching, and were followed for a mean of 4.5 years. Before treatment, the patients were classified into one of three clinical groups: (1) palpable sternomastoid tumor, (2) muscular torticollis (thickening and tightness of the sternocleidomastoid muscle), and (3) postural torticollis (torticollis but no tightness or tumor).

Results: Of the 821 patients, 452 (55%) had a sternomastoid tumor; 276 (34%), muscular torticollis; and ninety-three (11%), postural torticollis. Multivariate analysis of the outcomes showed that (1) the duration of treatment was significantly associated with the clinical group ($p < 0.0001$), a passive rotation deficit of the neck ($p < 0.0001$), involvement of the right side ($p < 0.0001$), difficulties with the birth ($p < 0.009$), and age at presentation ($p < 0.0001$); (2) the overall final assessment score was associated with the rotation deficit ($p = 0.02$), age at presentation ($p = 0.014$), and duration of treatment ($p < 0.0001$); and (3) subsequent surgical treatment was required by 8% (thirty-four) of the 452 patients in the sternomastoid tumor group compared with 3% (eight) of the 276 patients in the muscular torticollis group and 0% (none) of the ninety-three patients in the postural torticollis group.

Conclusions: This large prospective study demonstrated that controlled manual stretching is safe and effective in the treatment of congenital muscular torticollis when a patient is seen before the age of one year. The most important factors that predict the outcome of manual stretching are the clinical group, the initial deficit in rotation of the neck, and the age of the patient at presentation. Surgical treatment is indicated when a patient has undergone at least six months of controlled manual stretching and has residual head tilt, deficits of passive rotation and lateral bending of the neck of >15°, a tight muscular band or tumor, and a poor outcome according to our special assessment chart.

The reported prevalence of congenital muscular torticollis has ranged from 0.3% (fifteen of 5079 live births) to 2.0% (forty-three of 2190 live births)[1-7]. The typical lesion is a hard mass within the substance of a tight sternocleidomastoid muscle. It is often recognized when a child is one to four weeks old. The size of the lesion ranges from 1 to 3 cm in its largest transverse diameter. It is firm and smooth, movable beneath the skin, and apparently tender to touch[8]. After reaching its maximum size, the mass gradually recedes at varying rates within the first year and, in most patients, the muscle tightness subsides after the mass has resolved. In some patients, large portions of the muscle may become fibrotic. Secondary or associated skull and facial asymmetry or plagiocephaly are often found.

Macdonald[9] divided patients with congenital muscular torticollis into two groups: those with a sternomastoid tumor and those with tightness of the sternocleidomastoid but no clinical tumor, which he termed muscular torticollis. Postural torticollis is a term used to describe patients with congenital torticollis who have all the clinical features of torticollis but with no demonstrable tightness or tumor of the muscle[10]. However, a clear distinction has not been made between postural torticollis and congenital muscular torticollis in the literature, and in most series the term congenital muscular torticollis includes all three groups. To facilitate comparison with other series in the literature, we adopted these three terms (sternomastoid tumor, muscular torticollis, and postural torticollis) for our study.

Reports of the natural history and the outcome of treatment have varied considerably[4,9-13]. A common observation is that 54% (twenty-two of forty-one) to 70% (194 of 277) of the sternomastoid tumors resolve within the first year after birth[9,11]. Some patients have a degree of residual fibrosis of the muscle without an obvious clinical problem, and others (9% [six of sixty-six] to 21% [eighteen of eighty-four]) have progression to frank muscular torticollis and clinical deformity that requires surgical intervention[4,9,10,13]. Infants seen initially with muscular torticollis have an outcome similar to that in infants with a sternomastoid tumor[4].

Although it is well accepted that late cases of torticollis with a definite tight muscular band should be treated operatively, there is no clear consensus on the management of early cases. Treatment recommendations include observation only[14], application of an orthosis[11,15], an active home program of stimulation exercise and positioning[1,11,15], gentle manual stretching[1,4,11,14-17], vigorous manual manipulation with rupturing of the tight sternocleidomastoid muscle[10], and various types of operative procedures[2,4,8,18,19].

Despite the controversies, manual stretching is still the most common treatment for both sternomastoid tumor and muscular torticollis[2,10-12,14-18,20,21]. Success rates have ranged from 61% (nineteen of thirty-one)[14] to 99% (ninety-nine of 100)[15]. However, most of the studies have been retrospective, they often have consisted of small numbers of patients with a wide age-range, and data often have been collected over a long period of time. The type of congenital muscular torticollis has rarely been defined, the severity of disease prior to treatment has not been clear, the protocol and details of the stretching program have not been standardized, and no standard assessment methods have been used to compare the patients before and after treatment.

The main objective of the present prospective longitudinal study was to evaluate, according to a standard clinical classification system and assessment method, the effect of a standardized manual stretching program on a large group of patients with clearly defined congenital muscular torticollis. Another objective was to evaluate the factors that predict the outcome of treatment.

Materials and Methods

A total of 1086 patients with congenital muscular torticollis were first seen at birth or before the age of one year at our special Torticollis Clinic between 1985 and 1997. Of those patients, 821 were treated with a manual stretching program and were included in the study.

During the same period, 237 other patients who were seen in the Torticollis Clinic were not included in this study. Of those patients, 117 who had congenital muscular torticollis presented after the age of one year and another 120 patients had another type of torticollis such as acute torticollis, congenital anomalies of the cervical spine, spasmodic torticollis, obvious ocular torticollis, or another form of neurogenic or organic torticollis.

The information recorded for all patients seen in the clinic included gender, age at presentation, side of the torticollis, birth history and obstetrical data (breech presentation, vacuum extraction, forceps delivery, or caesarean section), and evidence of hip dysplasia and any other associated congenital anomalies.

Clinical Classification of Congenital Muscular Torticollis

All of the patients with congenital muscular torticollis were placed into one of the following three clinical groups[4,9,10]: (1) sternomastoid tumor group (those with a clinically palpable sternomastoid tumor), (2) muscular torticollis group (those with clinical thickening and tightness of the sternocleidomastoid muscle), and (3) postural torticollis group (those with postural head tilt and clinical features of torticollis but without tightness or tumor of the sternocleidomastoid muscle).

In the sternomastoid tumor group, the largest transverse diameter of the mass was measured to the nearest 0.5 cm. The distribution of the mass within the muscle was described as cephalad, middle, or caudad third or whole muscle-length involvement.

Subgroups According to Limitation of Passive Range of Rotation of the Neck

The range of rotation of the neck was measured with a specially designed arthrodial protractor. The infant's shoulders were stabilized in the supine position, and the head and neck were supported by the examiner in the neutral position, with regard to flexion and extension, over the edge of the examination table. This position allowed a detailed examination of the whole sternocleidomastoid muscle and at the same time allowed the neck to be rotated and moved freely in all directions. Normal infants have a passive range of rotation of up to 110° to each side. Clinical experience suggests that, compared with measurement of side-bending, measurement of rotation has better interexaminer reliability[1,11,14-16]. In a pilot study to evaluate the reproducibility of rotation measurements, we found an interexaminer correlation coefficient of 0.71 (unpublished data). The passive range of rotation of the neck on the side of the torticollis was compared with that on the normal side, and limitations were classified into two subgroups: a rotation deficit of ≤15° and a rotation deficit of >15°.

Age-Group

The age at presentation was subdivided into four groups: before the age of one month, at one to three months, at three to six months, and at six months to one year.

Treatment Protocol

We used a manual stretching program for all patients with clinical congenital muscular torticollis who presented with a demonstrable deficit of passive rotation of the neck of >10°. The hot, humid, and crowded local environment precluded

TABLE I Sheet for Scoring Results by Evaluation Category*

Category	Score 3 Points	2 Points	1 Point	0 Points
Rotation deficit *(deg)*	<5	6-10	11-15	>15
Lateral bending deficit *(deg)*	<5	6-10	11-15	>15
Craniofacial asymmetry	None	Mild	Moderate	Severe
Residual band (none, lateral, cleidal, or sternal)	None	Lateral	Lateral, cleidal	Cleidal, sternal
Head tilt (none, lateral, cleidal, or sternal)	None	Mild	Moderate	Severe
Subjective assessment by parents (cosmetic and functional)	Excellent	Good	Fair	Poor

*An overall score of 16, 17, or 18 points indicates an excellent result; 12 to 15 points, a good result; 6 to 11 points, a fair result; and <6 points, a poor result.

the use of a supportive collar or orthosis. Patients with a deficit of passive rotation of <10° were managed with an active home program of stimulation exercises and positioning that was taught to the parents.

Manual stretching was done three times a week by a properly trained and experienced physiotherapist who used a standardized program. Each session consisted of three repetitions of fifteen manual stretches of the tight muscle with a gentle force sustained for one second and a rest period of ten seconds in between. The parents were also taught to carry out a home program of active positioning with specific instructions that they should not do any passive stretching or manipulation. Treatment duration was defined as the time between the initial assessment and the time that full passive rotation of the neck was regained or when there was no further improvement after more than six months of treatment.

The indication for surgery was a persistent head tilt, with deficits of passive rotation and lateral bending of the neck of >15°, a tight band or tumor in the sternocleidomastoid, and a poor result according to the special assessment chart (Table I). Surgical candidates either had not responded or had shown no further improvement after at least six months of manual stretching. The majority of patients had a caudad unipolar open release with partial excision of the clavicular and sternal heads of the sternocleidomastoid. Postoperatively, after the wound had stabilized, an intensive physiotherapy program including scar massage, maintenance of full passive motion of the neck, and active strengthening exercises for a period of three to four months was prescribed. For patients who had a surgical release when they were more than two years old, a postoperative multiadjustable torticollis brace was prescribed for a period of three months, in addition to the physiotherapy program, to maintain the neck in the corrected position.

Follow-up Assessment

All patients were followed regularly in the special Torticollis Clinic; they were seen every two months during treatment and at regular intervals until the final assessment. The degree of head tilt, the active and passive range of rotation and lateral bending of the head, facial asymmetry, the size of the tumor and the time of disappearance of the tumor, and any complications such as ecchymosis or fracture of the clavicle were recorded at the time of the visit.

The outcome of treatment was analyzed with reference to the total duration of the treatment period, the overall score at the final assessment, and the need for operative treatment. The overall results were graded according to a scoring system and were designated as excellent, good, fair, or poor on the basis of both subjective and objective criteria derived from a number of previous studies (Table I)[11,14,15,17,19].

Statistical Analysis

Both univariate and multivariate analysis were used to assess the data. Univariate analysis included the chi-square or chi-square exact test, t test, Mann-Whitney test, and Kruskal-Wallis test, where appropriate. Subgroup analyses were performed if there was a significant association between the two factors. The corresponding p values were adjusted by the number of paired comparisons performed. Multivariate analysis included generalized linear models and logistic regression with relative risks, and their exact 95% confidence intervals were calculated. For statistical analysis, the overall scores were subgrouped into an excellent or good group and a fair or poor group. Similarly, the rotation groups, consisting of the patients who had ≤15° of rotation and those who had >15°, were analyzed separately. The rotation groups and the clinical groups could not be fitted simultaneously into the same multivariate analysis; hence, these two variables were analyzed separately.

Statistical analyses were done with the SPSS for Windows statistical software (release 8.0; SPSS, Chicago, Illinois), S-PLUS (version 4.3; MathSoft, Seattle, Washington), and StatXact (version 2.05; CYTEL Software, Cambridge, Massachusetts) computer programs. The level of significance was set at 5%.

TABLE II Comparison of Age at Presentation and Clinical Group*

Clinical Group	Age at Presentation <1 Mo	1-3 Mo	3-6 Mo	6-12 Mo
Sternomastoid tumor (n = 452)	155 (34%)	264 (58%)	28 (6%)	5 (1%)
Muscular torticollis (n = 276)	34 (12%)	85 (31%)	107 (39%)	50 (18%)
Postural torticollis (n = 93)	4 (4%)	14 (15%)	52 (56%)	23 (25%)
Total (n = 821)	193 (24%)	363 (44%)	187 (23%)	78 (10%)

*The values are given as the number of patients.

Results

A total of 1086 patients with congenital muscular torticollis who were less than one year old were seen from 1985 to 1997. Of those patients, 821 were treated with the manual stretching program and 788 (96%) of them were available for the overall final evaluation. The mean duration of follow-up was 4.5 years (range,1.5 to 13.0 years).

Clinical Classification

A total of 452 patients (55%) had a sternomastoid tumor, 276 (34%) had muscular torticollis, and ninety-three (11%) had postural torticollis. Torticollis was found on the left side in 51% (420) of the patients and on the right side in 49% (401), with no significant difference between the numbers with right and left involvement in any of the three clinical groups. The male:female ratio was 3:2 in all three clinical groups.

Age at Presentation

Of the total of 821 patients, 193 (24%) presented to the clinic within one month after birth; 363 (44%), from one to three months; 187 (23%), from three to six months; and seventy-eight (10%), from six to twelve months (Table II).

One hundred and fifty-five (34%) of the 452 patients who had a sternomastoid tumor were seen within one month after birth, and 264 (58%) were seen from one to three months after birth. Only thirty-four (12%) of the 276 patients with muscular torticollis presented in the first month, whereas eighty-five (31%) were first seen from one to three months after birth. Of the ninety-three patients who had postural torticollis, seventy-five (81%) presented more than three months after birth. A significant difference with regard to the age at presentation was detected between the sternomastoid tumor group and the muscular torticollis group ($p < 0.0001$), the sternomastoid tumor group and the postural torticollis group ($p < 0.001$), and the muscular torticollis group and the postural torticollis group ($p < 0.003$).

The mean age (and standard deviation) at presentation was 44 ± 33 days in the sternomastoid tumor group, 109 ± 74 days in the muscular torticollis group, and 143 ± 75 days in the postural torticollis group, indicating that the patients in the sternomastoid tumor group presented earlier than those in the other two groups ($p < 0.001$).

Limitation of Range of Rotation of the Neck

A rotation limitation of >15° was more prevalent in the sternomastoid tumor group (72% [327] of 452 patients) than in the muscular torticollis group (32% [eighty-eight] of 276 patients) ($p < 0.0001$) or the postural torticollis group (4% [four] of ninety-three patients) ($p < 0.0001$) (Table III).

The 419 patients with a rotation deficit of >15° had a higher rate of breech presentation (17% [seventy-two], $p < 0.0001$) and vacuum extraction (30% [125], $p < 0.0001$) than the 402 patients who had a rotation deficit of ≤15° (7% [thirty] and 14% [fifty-six], respectively). Seven (2%) of the 402 patients with a rotation deficit of ≤15° had hip dysplasia compared with 7% (thirty) of the 419 who had a rotation deficit of >15° ($p < 0.0002$). The median age at presentation of the patients with a rotation deficit of >15° (thirty-six days) was earlier than that of the patients with a rotation deficit of ≤15° (ninety-five days) ($p < 0.0001$).

TABLE III Data on Clinical Groups in Terms of Rotation Deficit and Outcome

Clinical Group (N = 821)	Rotation Deficit* ≤15°	Rotation Deficit* >15°	Median Duration of Treatment *(mo)*	Overall Score* Excellent or Good	Overall Score* Fair or Poor	Surgical Treatment*
Sternomastoid tumor (n = 452)	27.7%	72.3%	3.7	87.8%	12.2%	7.5%
Muscular torticollis (n = 276)	68.1%	31.9%	2.5	93.8%	6.2%	3.1%
Postural torticollis (n = 93)	95.7%	4.3%	1.4	98.9%	1.1%	0.0%

*The values are given as the percentage of patients.

TABLE IV Significant Risk Factors Associated with Overall Final Score* (N = 788)

	Rating of Overall Final Score†		
	Excellent or Good (N = 717)	Fair or Poor (N = 71)	P Value‡
Clinical group			
Postural torticollis	86 (12.0%)	1 (1.40%)	0.0008§
Muscular torticollis	242 (33.8%)	16 (22.5%)	
Sternomastoid tumor	389 (54.3%)	54 (76.1%)	
Rotation deficit			
≤15°	363 (50.6%)	16 (22.5%)	<0.0001
>15°	354 (49.4%)	55 (77.5%)	
Age at presentation			
<29 days	178 (24.8%)	11 (15.5%)	<0.0001
29-90 days	301 (42.0%)	48 (67.6%)	
91-365 days	238 (33.2%)	12 (16.9%)	
Side of involvement#			
Left	376 (52.5%)	28 (39.4%)	0.0350
Right	340 (47.5%)	43 (60.6%)	
Difficulties with birth			
Yes	328 (45.7%)	43 (60.6%)	0.0170
No	389 (54.3%)	28 (39.4%)	
Duration of treatment *(mo)***	4.3 (0.90)	5.0 (0.90)	<0.0001††

*Data are given for 788 patients as thirty-three had missing information. †The values are given as the number of patients. ‡As determined with the chi-square test. §As determined with the chi-square exact test. #Data are given for 787 patients as one patient had missing information. **Logarithmic transformation was applied to make the variable less skewed. The values are given as the mean (and the standard deviation). ††As determined with the t test.

Complications of Manual Stretching

Sudden giving-way or snapping of the sternomastoid was observed during treatment with manual stretching in 8% (thirty-seven) of the 452 patients with a sternomastoid tumor. Stepwise logistic regression revealed that those who had hip dysplasia, left-sided involvement, a rotation deficit of >15°, and were less than one month old at presentation were at higher risk for this event. Sudden giving-way was followed by clinical signs of bruising and an increase in the range of motion of the neck, signifying a possible tear or rupture of the muscle. On subsequent follow-up examination at a mean of 4.5 years, sudden giving-way did not seem to have resulted in an increased need for operative treatment (two [5%] of the thirty-seven patients who had had giving-way had an operation compared with thirty-two [8%] of the 415 patients who had not had giving-way; p = 1.00). Overall, the clinical result was good or excellent in thirty-five (95%) of the thirty-seven patients who had giving-way and in 354 (85%) of the 415 pa-

TABLE V Multivariate Logistic Model for Final Overall Score*

	Relative Risk (95% Confidence Interval)	P Value
Patients with rotation deficit of >15°	2.17 (1.13, 4.17)	0.020
Age at presentation†		
29-90 days	2.50 (1.21, 5.17)	0.014
91-365 days	1.42 (0.55, 3.70)	0.471
Duration of treatment‡ *(mo)*	2.19 (1.49, 3.21)	<0.0001

*Data are given for 788 patients as thirty-three had missing information. †Patients who were less than twenty-nine days old at the time of presentation were considered the reference group. ‡Logarithmic transformation was applied to make the variable less skewed.

TABLE VI Significant Risk Factors for Predicting the Necessity of Surgical Treatment (N = 821)

	Rating of Overall Final Score* (*no. of patients*)		
	Excellent or Good (N = 779)	Fair or Poor (N = 42)	P Value*
Clinical group			
Postural torticollis	93 (11.9%)	0 (0.0%)	0.0018†
Muscular torticollis	268 (34.4%)	8 (19.0%)	
Sternomastoid tumor	418 (53.7%)	34 (81.0%)	
Rotation deficit			
≤15°	397 (51.0%)	5 (11.9%)	<0.0001
>15°	382 (49.0%)	37 (88.1%)	
Age at presentation			
<29 days	188 (24.1%)	5 (11.9%)	0.0010
29-90 days	333 (42.7%)	30 (71.4%)	
91-365 days	258 (33.1%)	7 (16.7%)	
Lateral bending deficit			
<5° or 6-10°	461 (59.2%)	11 (26.2%)	<0.0001
11-15° or >15°	318 (40.8%)	31 (73.8%)	
Craniofacial asymmetry‡			
Normal or mild	579 (75.5%)	25 (59.5%)	0.0210
Moderate or severe	188 (24.5%)	17 (40.5%)	
Head tilt§			
Normal or mild	709 (91.2%)	7 (16.7%)	<0.0001
Moderate or severe	68 (8.8%)	35 (83.3%)	
Overall final score#			
Excellent or good	717 (96.1%)	0 (0.0%)	<0.001†
Fair or poor	29 (3.9%)	42 (100%)	

*As determined with the chi-square test. †As determined with the chi-square exact test. ‡Data are given for 809 patients as twelve had missing information. §Data are given for 819 patients as two had missing information. #Data are given for 788 patients as thirty-three had missing information.

tients who had not had giving-way ($p = 0.291$).

Determinants and Predictors of Outcome of Treatment

The outcome of treatment was analyzed with respect to the duration of treatment, the overall score at the final assessment, and the necessity for operative treatment.

Duration of Treatment

The median duration of treatment was 3.7 months for the sternomastoid tumor group, 2.5 months for the muscular torticollis group, and 1.4 months for the postural torticollis group. A significant difference with respect to duration of treatment was detected between the sternomastoid tumor group and the muscular torticollis group, the sternomastoid tumor group and the postural torticollis group, and the muscular torticollis group and the postural torticollis group ($p < 0.0001$ for each) (Table III).

The duration of treatment was found, with univariate analysis, to be significantly associated with the age at presentation, the clinical group, the initial rotation deficit, breech presentation, difficulties with the birth, and the side of torticollis ($p < 0.05$). Thus, a patient with a sternomastoid tumor of the right side, whose birth was associated with difficulties, who is seen more than one month after birth, and who has a rotation deficit of >15° is more likely to need longer treatment.

Multivariate analysis was performed to identify the most important determinants or predictors of the duration of treatment. The significant variables from the univariate analysis were entered into the generalized linear model analysis to assess the confounding factors associated with duration of treatment. Logarithm transformation was applied to make the variable less skewed. The results showed that the duration of treatment was significantly associated with a rotation deficit of >15° ($p < 0.0001$), the clinical group ($p < 0.0001$), an age of more than one month at presentation ($p < 0.0001$), involvement of the right side ($p < 0.0001$), and difficulties with the birth ($p < 0.009$).

Overall Final Score

Of the 821 patients in our study, 788 (96%) were available for the overall final evaluation at a mean of 4.5 years (range, 1.5 to 13.0 years). The results were significantly different among the clinical groups. One percent (one) of the eighty-seven patients in the postural torticollis group, 6% (sixteen) of the 258 in the muscular torticollis group, and 12% (fifty-four) of the 443 in the sternomastoid tumor group had a fair or poor result. A significant difference was detected between the sternomastoid tumor group and the muscular torticollis group ($p = 0.033$) and between the sternomastoid tumor group and the postural torticollis group ($p = 0.006$) but not between the muscular torticollis group and the postural torticollis group ($p = 0.249$).

With use of univariate analysis, the final overall score was found to be strongly associated with the clinical group ($p = 0.0008$), rotation deficit ($p < 0.0001$), age at presentation ($p < 0.0001$), side of involvement ($p = 0.035$), and difficulties with the birth ($p < 0.017$). Thus, a patient with a sternomastoid tumor, whose birth was associated with difficulties, who is first seen more than one month after birth, and who has a rotation deficit of >15° is more likely to have a worse final overall score (Table IV).

Multivariate analysis with use of the stepwise logistic regression analysis showed that worse overall scores were associated with a rotation deficit of >15° ($p = 0.02$), an age of one to three months at the time of presentation ($p = 0.014$), and the duration of treatment ($p < 0.0001$) (Table V). After adjustment, the side of involvement and the clinical group were not found to be associated with worse overall scores.

Prediction of the Need for Surgical Treatment

Thirty-four (8%) of the 452 patients who had a sternomastoid tumor needed an operation compared with eight (3%) of the 276 patients in the muscular torticollis group. None of the patients who had postural torticollis had surgical treatment (Table III). A significant difference was detected between the sternomastoid tumor group and the muscular torticollis group ($p = 0.027$) and between the sternomastoid tumor group and the postural torticollis group ($p = 0.006$) with respect to the need for an operation. No significant difference was found between the muscular torticollis group and the postural torticollis group ($p = 0.63$).

Univariate analysis showed that the factors significantly associated with operative treatment were the age at presentation, rotation deficit, clinical group, lateral bending, craniofacial asymmetry and head tilt, and overall score (Table VI). Thus, a patient with a sternomastoid tumor who has craniofacial asymmetry and head tilt, is first seen more than one month after birth, and has a rotation deformity of >15° and a poor overall score is more likely to need surgical treatment. No significant association was found between the side of involvement and the need for an operation ($p = 0.081$).

Multivariate analysis with the stepwise logistic regression model showed that the confounding risk factors for an operation were a late age at presentation ($p = 0.008$) and the clinical group (sternomastoid tumor) ($p = 0.023$) or a rotation deficit of > 15° ($p < 0.0001$) (Table VII).

Discussion

It is difficult to find reliable reports on the outcome of treatment of congenital muscular torticollis. McDaniel et al.[13], in their comprehensive review of the findings in patients with a sternomastoid tumor, reported that 30% (eighty-three of 277) to 46% (nineteen of forty-one)[9,11] had natural progression to simple fibrosis of the sternocleidomastoid without substantial clinical problems and that the percentage of patients who had progression to frank torticollis requiring surgical treatment ranged from 9% (six of sixty-six) to 21% (eighteen of eight-four)[4,9,10,12,13]. Hulbert[10] was the first to describe clearly the importance of differentiating postural torticollis from true sternomastoid tumor and muscular torticollis. The natural history of postural torticollis is always benign; it resolves within a few months, and operative treatment is never indicated. Jones[12] prospectively studied 100 patients with congenital muscular torticollis and followed them until they were twelve years old. Sixty-six had a sternomastoid tumor, and thirty-four had muscular torticollis. Fifty percent of the patients with a sternomastoid tumor were found to have no evidence of the disorder at six months of age, 30% had some residual tightness, and 9% required operative treatment of

TABLE VII Two Possible Multivariate Logistic Models for Predicting the Necessity of Surgical Treatment

	Model I		Model II	
	Relative Risk (95% Confidence Interval)	P Value	Relative Risk (95% Confidence Interval)	P Value
Clinical Group*				
Sternomastoid tumor	2.89 (1.16, 7.21)	0.023		
Rotation deficit of >15°			8.02 (2.97, 21.72)	<0.0001
Age at presentation†				
29-90 days	3.66 (1.39, 9.62)	0.008	3.99 (1.51, 10.52)	0.005
91-365 days	2.72 (0.76, 9.81)	0.125	2.60 (0.77, 8.76)	0.122

*Ninety-three patients in the postural torticollis group were excluded because none needed surgical intervention. †Patients who were less than twenty-nine days old at the time of presentation were used as the reference group.

the torticollis. Of the thirty-four patients with muscular torticollis, four (12%) had an operation and eight (24%) had residual fibrosis.

Emery[15] reported on a series of 100 patients with congenital muscular torticollis who presented before the age of two years (range, 0.5 to 15.5 months) and were treated with a defined home protocol of stretching and active positioning stimulation. A successful end point was considered to be a lateral bending or rotation deficit of <6°. The duration of treatment was used as the outcome dependent variable. The mean duration of treatment was 6.9 months for the sternomastoid tumor group and 3.9 months for the muscular torticollis group. According to multiple regression analysis, the only significant determinant or predictor of treatment duration was the severity of restriction of neck rotation at the beginning of treatment ($p = 0.0074$). A high attrition rate occurred in that study, with only 100 of 181 patients available for the final analysis. Only the duration of treatment was used as an outcome parameter, and the follow-up period was relatively short. The treatment program was performed by the parents and not by trained therapists, making standardization of the technique difficult.

To our knowledge, the present study is the only prospective series with standardized pretreatment classification of the clinical groups, validated assessment techniques, and a standard manual stretching protocol performed by experienced physiotherapists. To identify the determinants of the outcome of treatment in more detail, three parameters were measured in this study: the duration of treatment, the overall clinical result, and the necessity for operative treatment.

The mean duration of treatment was found to be significantly longer in the sternomastoid tumor group (3.7 months) than in the muscular torticollis group (2.5 months) ($p < 0.0001$). Univariate analysis demonstrated that the clinical group (sternomastoid tumor), an older age at presentation, difficulties with the birth, involvement of the right side, and rotation deformity of >15° were all significantly associated with a longer duration of treatment ($p < 0.05$). The clinical group (sternomastoid tumor), age at presentation, more severe rotation deformity, and duration of treatment all were predictors of the final score.

The current study is unique in that the necessity for operative treatment was used as an outcome parameter. All of the patients had regular follow-up examinations in a special torticollis clinic with use of a clear assessment protocol. The follow-up rate was 96%. Only patients with a fair or poor final overall score were treated operatively. Statistical analysis showed that the most important predictors of the need for operative treatment were the clinical group (sternomastoid tumor), the severity of the rotation deformity, and an older age at presentation. None of the patients in the postural torticollis group needed surgical intervention, an observation that is consistent with the findings of Hulbert[10], who demonstrated the importance of recognizing postural torticollis.

The current series of patients had much better overall results with a lower rate of operative intervention compared with those in most of the reported series[2,4,10-12,14-18,20,21]. If patients with postural torticollis were included in the analysis, the success rate would have been even higher. Several factors may explain the high success rate associated with manual stretching in our series. First, all manual stretching was conducted with use of a clear protocol by trained physiotherapists rather than by parents. Second, the majority of our patients were first seen early, in the first few months of life, and treatment was started relatively early.

The effect of stretching on muscles is not totally understood. A recent study on surgical specimens of sternomastoid tumor, with use of detailed electron microscopic and immunohistochemical techniques, showed that myoblasts exist in the proliferating interstitium of the tumor and are primarily responsible for the maturation and resolution of the tumor, possibly by producing normal myofibrils[22]. Given the right stimulus and a favorable environment, these myoblasts may be activated and contribute to the regeneration and repair of the abnormal sternocleidomastoid. Controlled manual stretching may provide a favorable stimulation for myogenesis. In contrast, without the right stimulus or in cases of severe damage, fibroblasts may prevail with resultant progressive fibrosis, as is typical of late muscular torticollis.

The present study showed that controlled manual stretching 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 that predict the outcome of manual stretching are the clinical group (sternomastoid tumor, muscular torticollis, or postural torticollis), the initial deficit in rotation of the neck, and the age at presentation. Surgical treatment is indicated when a patient has undergone at least six months of controlled manual stretching and has a residual head tilt, deficits of passive rotation and lateral bending of the neck of >15°, a tight band or tumor, and a poor result defined as an overall score of <6 points according to our special assessment scale. ■

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