The Clinical Presentation and Outcome of Treatment of Congenital Muscular Torticollis in Infants—A Study of 1,086 Cases

By J.C.Y. Cheng, S.P. Tang, T.M.K. Chen, M.W.N. Wong, and E.M.C. Wong
Hong Kong

Background/Purpose: The main objectives of this study were to define the clinical patterns and characteristics of congenital muscular torticollis (CMT) presented in the first year of life and to study the outcome of different treatment methods.

Methods: This is a prospective study of all CMT patients seen in 1 center over a 12-year period with uniform recording system, assessment methods, and treatment protocol.

Results: From a total of 1,086 CMT infants, 3 clinical subgroups of sternomastoid tumor (SMT; 42.7%), muscular torticollis (MT; 30.6%), and postural torticollis (POST; 22.1%) were identified. The SMT group was found to present earlier within the first 3 months and was associated with higher incidence of breech presentation (19.5%), difficult labor (56%), and hip dysplasia (6.81%). Severity of limitation of passive neck rotation range (ROTGp) was found to correlate significantly with the presence of SMT, bigger tumor size, hip dysplasia, degree of head tilt, and craniofacial asymmetry.

Conclusions: A total of 24.5% of the patients with initial deficits of passive rotation of less than 10° showed excellent and good outcome with active home positioning and stimulation program. The remaining cases with rotation deficits of over 10° and treated with manual stretching program showed an overall excellent to good results in 91.1% with 5.1% requiring subsequent surgical treatment. The most important prognostic factors for the necessity of surgical treatment were the clinical subgroup, the ROTGp, and the age at presentation ($P < .001$).

INDEX WORDS: Torticollis, outcome.

TORTICOLLIS in Latin means twisted neck and was first defined by Tubby in 1912 as "A deformity, either congenital or acquired, characterized by lateral inclination of the head to the shoulder, with torsion of the neck and deviation of the face."[1] The term *congenital muscular torticollis* has been used by various investigators[2-8] to denote a neck deformity primarily involving shortening of the sternomastoid muscle that is detected at birth or shortly after birth. This clearly should be differentiated from many other congenital and acquired types of torticollis such as congenital cervical vertebral anomalies, posttraumatic infections and inflammation of adjacent structures, neoplastic conditions, and miscellaneous types of structural and functional neurological causes. In infants with torticollis, the head typically is tilted toward the side of the affected muscle and rotated toward the opposite side. In many cases, a mass or tumor can be palpated in the involved muscle. Skull and facial asymmetry or plagiocephaly also may be present. Although a combination of various theories has been proposed, the true etiology of torticollis remains uncertain. Among them is birth trauma, which proposed that congenitally shortened sternomastoid muscle was torn at birth with the formation of a hematoma, which then underwent fibrous contracture.[9] The ischemic hypothesis postulated that venous occlusion produced ischemia in the sternomastoid muscle.[10] Intrauterine malposition was considered a possible etiology.[3,11] Other hypotheses are the hereditary hypothesis, neurogenic theory, and the infection theory.[7,12] One of the latest hypotheses proposed that the condition could be the sequel of an intrauterine or perinatal compartment syndrome.[13]

Macdonald further divided the congenital muscular torticollis (CMT) into sternomastoid tumor group (SMT) and those with tightness of the sternocleidomastoid muscle (SCM) but no clinical "tumor" as muscular torticollis (MT).[8] Postural torticollis (POST) was used to describe those congenital torticollis with all the clinical features of torticollis but with no demonstrable tightness nor tumor of the sternomastoid muscle.[6] However, such distinction of postural torticollis from the CMT was not made clearly in the literature and in most series the term *CMT* would include all these 3 groups. To facilitate

From the Department of Orthopaedics & Traumatology, Centre for Clinical Trials & Epidemiological Research, The Chinese University of Hong Kong, Shatin, NT, Hong Kong; the Division of Orthopaedic Surgery, Children's Hospital, Chongqing University of Medical Sciences, China; and Physiotherapy Department, Kowloon Hospital, Hong Kong.

Address reprint requests to Jack C.Y. Cheng, MD, FRCSEd (Orth), Department of Orthopaedics & Traumatology, The Chinese University of Hong Kong, 5th Floor, Clinical Sciences Building, Prince of Wales Hospital, Shatin, NT, Hong Kong.

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comparison with the literature, such synonymous description was adopted in this study.

The reported incidence of torticollis varied from 0.3% to 1.9%.[1,4,5,14-16] Treatment includes observation, application of orthosis, active home exercise program, gentle manual stretching, vigorous manual myotomy, and various types of surgical procedures. There are a number of inherent problems when one reviews the literature of the past 100 years. Most series are historical series based on small numbers over long period of time. Most studies include mixed clinical group of early and late presentation cases without clear indications for treatment nor standard subjective nor objective assessment methods. As a result, the conclusions based on the condition and the outcome of treatment are not comparable among the different series.

The current study is a prospective study of all CMT patients seen in 1 center over a 12-year period with a uniform recording system and assessment methodology. This study is an extension of 1 of our previous studies[14] with larger number of patients, many longer follow-up periods, and more detailed outcome analysis with a special scoring system. The main objectives were to define the clinical patterns and characteristics of CMT in infants presenting in the first year of life using standard grouping and assessment methods and to use this as a basis to study the outcome of a well-defined treatment protocol.

MATERIALS AND METHODS

All patients less than 1 year of age with torticollis treated in the special Children's Torticollis Clinic center from 1985 to 1997 were included in the study. Patients with acute torticollis, congenital anomalies of the cervical spine, spasmodic torticollis, and other forms of neurogenic, ocular, and organic torticollis were excluded from the study. The following information was recorded: sex, the age at presentation, the side of the torticollis, birth history and obstetric data, presence of hip dysplasia, associated spinal and musculoskeletal anomalies, the presence of head tilt and craniofacial asymmetry, the limitation of range of motion of the neck in rotation, and side flexion as compared with the normal side. All the data were analyzed independently by one of the authors.

Clinical Groups of Congenital Muscular Torticollis

All the cases of congenital torticollis were subdivided into the following 3 clinical groups: (1) SMT group with definite presence of clinically palpable sternomastoid "tumor," (2) MT group—muscular torticollis group without palpable or visible tumor but with clinical thickening or tightness of the sternomastoid muscle on the affected side, and (3) POST group—a third group without features of the above 2 groups; most patients in this group had postural head tilt or late-onset ocular torticollis.

Measurement of Limitation of Range of Motion of the Neck

Range of motion of the neck was measured using the arthrodial protractor with the baby or child in the 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 is free to rotate and move in all directions (Fig 1). This position also would allow a detailed examination of the neck and the whole SM muscle from the origin to the clavicular and sternal insertion. Clinical experience suggested that the rotation element (ROT) is easier to measure with good interexaminer reliability and is thus preferred by therapists to side flexion.[14,17] The passive range of motion of neck were assessed and compared with the normal side. The limitations in passive ROT were recorded for data analysis and were subclassified into 4 subgroups according to the severity of passive neck ROT deficits: ROTGp I, no actual ROT limitation; ROTGp II, ROT limitation of less than or equal to 15°; ROTGp III, ROT limitation of 16° to 30°; ROTGp IV, ROT limitation of more than 30°.

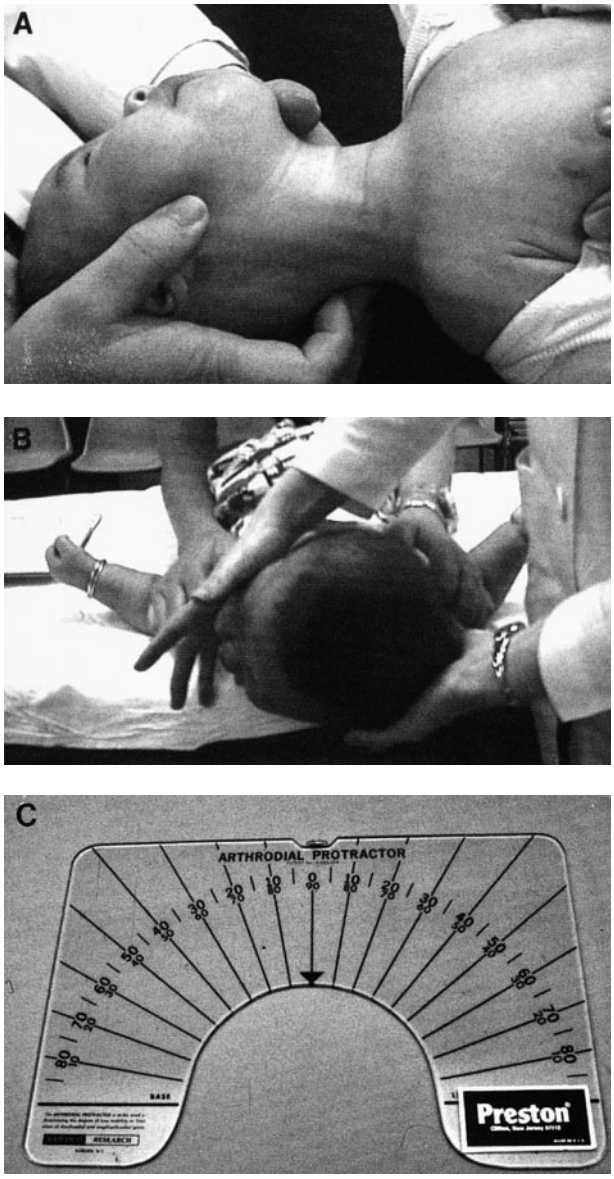

Fig 1. (A,B) Proper positioning of the neck for examination and measurement of passive rotation. (C) Arthrodial protractor.

Age Groups

For demographic and epidemiological study, the time of presentation of all the patients were grouped into those presenting before the age of 1 month, from 1 to 3 months, 3 to 6 months, and 6 months to 1 year.

Treatment Protocol

Home treatment group. For cases of minimum deficits of passive rotation of less than 10°, an active home stimulation exercise and positioning program were used. A standard brochure and a set of slides to illustrate the home program were used to enhance the instructions and training of the caretaker. The accuracy of the home exercises was confirmed and checked by the author at regular intervals. The end point of treatment was the attainment of full or less than 5° of limitation of passive rotation of the neck.

Manual stretching group. The indications for manual stretching are for patients who have obvious CMT and are less than 1 year old with deficits of passive rotation of the neck of more than 10° irrespective of the clinical groups.

Manual stretching was done by properly trained and experienced physiotherapists with a standardized program 3 times per week. 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 shown after more than 6 months of treatment.

Surgical treatment group. The indications for surgery were for patients with significant head tilt and deficits of passive rotation and side flexion of the neck greater than 10° to 15° and the presence of tight band or tumor in the SCM. They either have not responded to or improved additionally after at least 6 months of physiotherapy manual stretching. For the majority of patients a uniform method of distal unipolar open release and partial excision of the clavicular and sternal heads of the sternomastoid muscle was done. Postoperatively, an intensive program of physiotherapy was prescribed that included scar treatment, maintenance of full passive range of motion of the neck, and active strengthening exercise for a period of 3 to 4 months.

Follow-Up Assessments

All patients were followed up regularly in the Torticollis Clinic with detail documentation of the head tilt, active and passive range of motion of rotation and side flexion of the neck, facial asymmetry, size of the tumor and time of disappearance of tumor, and treatment duration. For the operated group, the details of the operations, postoperative progress, and complications were noted. At the final assessment the overall results were graded by a scoring system based on both subjective and objective criteria and grouped as excellent, good, fair, and poor, respectively (Table 1). In this study the subjective score was based on interviewing the parents at the final assessment and inquiring about the overall cosmetic and functional results of the patient by an independent observer.

Statistical Analysis

Chi-square tests and Kruskal-Wallis test were used to assess possible association between the variables observed, in particular for patients with various degrees of ROT limitation in neck range, the different age presentation groups, and the different clinical groups of SMT, MT, and POST. Multivariate stepwise logistic regression was used to assess the confounding risk factors for operation. SPSS for Windows (Release 9.0, SPSS Inc, Chicago, IL), and StatXact (Version 2.05, CYTEL Software Corporation, Cambridge, MA) statistical software were used in the analyses. The level of significance was set at 5% in all comparisons, and all statistical testing was 2 sided.

RESULTS

From a total of 1,203 patients evaluated and followed up in the period of 1985 to 1997 with a mean follow-up of 3.5 years (range follow up of 1.5 to 13 years), a total of 1,086 cases seen before the age of 1 year were included in this study.

General Overall Clinical Findings

From the total number of 1,086 cases there were 434 girls and 652 boys with a ratio of 2:3. The mean gestation period was 39 weeks with a SD of 1.76 weeks. The mean birth weight was 3.2 kg ranging from 1.2 to 4.5 kg.

A total of 119 cases (13.00%) had breech presentation and another 15 (1.6%) with presentations other than that of the usual vertex presentation. The mode of delivery was summarized in Table 2.

Hip dysplasia diagnosed clinically and confirmed ultrasonographically was found in 4.1% of patients. Congenital spinal anomalies were found in 0.2%, and other musculoskeletal anomalies including varus toes, metatarsus adductus, postural and structural talipes equinovarus, and calcaneal valgus foot were found in 6.5% of all cases.

Craniofacial asymmetry of various degrees (mild, moderate, and severe) was found in 90.1% of patients (65% mild and 23.4% moderate) at first presentation. Clinically, they presented commonly with flattening of the occiput contralaterally and depression of the malar prominence ipsilaterally. Those with severe craniofacial asymmetry also would have downward displacement of

Table 1. Scoring Sheet for Overall Results

Overall Results	Excellent (3 points)	Good (2 points)	Fair (1 point)	Poor (0 points)
ROT deficits (degrees)	<5	6-10	11-15	>15
Side flexion deficits (degrees)	<5	6-10	11-15	>15
Craniofacial asymmetry	None	Mild	Moderate	Severe
Residual band (no, lateral, cleido, sternal)	None	Lateral	Lateral, cleido	Cleido, sternal
Head tilt (no, mild, moderate, severe)	None	Mild	Moderate	Severe
Subjective assessment by parents (cosmetic and functional)	Excellent	Good	Fair	Poor
Overall scores	16-18	12-15	6-11	<6

Table 2. Clinical Groups Versus Mode of Delivery and Age at Presentation

	SMT	MT	POST
No. of subjects (%)	515 (47.2)	334 (30.6)	241 (22.1)
Mode of Delivery (%)			
Vacuum	31.2	19.5	11
Cesarean section	17.7	20.5	24.2
Forcep	6.2	4.6	4.9
Age at Presentation (%)			
<1 mo	36.1	12	2.9
1-3 mo	56.5	33.5	14.1
3-6 mo	6.4	38.3	57.7
6-12 mo	1	16.2	25.3

the ear, eye, and mouth on the affected side. Head tilt was observed in all patients.

Clinical Groups

There were 515 cases of SMT (47.2%), 334 MT (30.6%), and 241 (22.1%) POST cases (Table 2). The torticollis occurred in the left side in 53% and right side in 47.0% with no statistical significant difference in all the 3 groups.

Breech presentation occurred in 19.5% of the SMT group, significantly higher than the other groups (χ^2 Exact test, $P < .001$; Table 2). For the mode of delivery, the SMT group differed significantly from the other 2 groups in having a much higher rate of forceps delivery and vacuum extraction (χ^2 Exact test, $P < .001$; Table 2).

Hip dysplasia was found in 6.8% of the SMT group and only in 1.9% of the MT group and 0.9% of the POST group (χ^2 Exact test, $P < .001$). Head tilt, craniofacial asymmetry also were present in a much higher rate and of more severe nature in the SMT group when compared with the other 2 groups.

For the SMT group the "tumor" was found clinically in the lower third of the sternomastoid muscle in 35%, middle third in 40.4%, upper third in 11.9% and over the whole muscle in 12.6%. The size of the sternomastoid tumor ranged from less than 1 cm (26.3%) to 4 cm in diameter with over 70% bigger than 2 cm.

Age at Presentation

From the 1,086 cases, 21.4% presented to the clinic within 1 month after birth and another 40.1% from 1 to 3 months. On more detailed breakdown, the SMT cases presented much earlier (Table 2). The differences between the 3 clinical groups were statistically significant (χ^2 Exact test, $P < .001$). The mean age of presentation was found to be 43.8 days in the SMT group, 106 days in MT, and 149 days in POST group indicating that the SMT group was found earlier than the other 2 groups (Kruskal-Wallis test, $P < .001$).

Limitation of Range of Rotation of the Neck

Of the 4 ROT groups, the most severe group, ROTGp IV, ie, with rotation limitation of over 30°, 95.8% were found in the SMT group (Table 3).

The ROTGp III and IV also were found to be associated with higher rate of breech presentation, assisted deliveries, and hip dysplasia (Table 3). All these associations were statistically significant (χ^2 Exact test, $P < .001$). The age at presentation was found to be earlier in the ROTGp III and IV cases. There also was a statistically significant correlation between the ROTGp III and IV with the severity of head tilt and craniofacial asymmetry.

Results of Treatment

According to the criteria and treatment protocol, 266 patients (24.5%) received active treatment at home, and 820 patients (75.5%) received manual stretching therapy. The mean follow-up of the patients was 4.5 years with a range from 1.5 years to 13 years.

Of the 266 cases with minimal limitation of rotation and side flexion (≤10°) and treated with active treatment program at home, no deterioration was detected. The breakdown of the patients showed that 24.2% belonged to the SMT group, 17.6% to the MT group, and 58.1% to the POST group. Of all the cases, 5% were changed to the manual stretching group when no improvement was seen within 4 weeks after the start of treatment. They all had excellent results at the final assessment. No patient required surgical intervention at the final assessment.

The majority of the 820 cases treated with manual stretching program belonged to the SMT group (55.1%), followed by MT (33.6%) and POST (11.3%). Most cases treated with manual stretching showed progressive improvement of the range of side flexion, rotation, the head tilt, the craniofacial asymmetry, and a decrease in the tumor size in the SMT group. The overall mean duration of treatment with physiotherapy was 118 days (3.9 months). The differences of duration of treatment between the different ROT groups were significant (Kruskal-Wallis test, $P < .001$; Fig 2).

The overall final outcome scores for the manual stretch group were excellent and good in 91.1%. Additional breakdown of the results showed that the percentage of fair to poor results was 1% in the POST group, 6.2% in

Table 3. ROT Grouping Versus Clinical Groups and Percentage of Hip Dysplasia

	SMT (%)	MT (%)	POST (%)	Hip Dysplasia (%)
ROTGp I (0°)	31 (6.1)	89 (26.7)	158 (65.6)	0
ROTGp II (<15°)	136 (26.7)	147 (44)	77 (32)	2.94
ROTGp III (16-30°)	233 (45.3)	93 (27.8)	6 (2.5)	5.94
ROTGp IV (>30°)	114 (22.2)	5 (1.5)	0 (0)	10.92
Total	(100)	(100)	(100)	

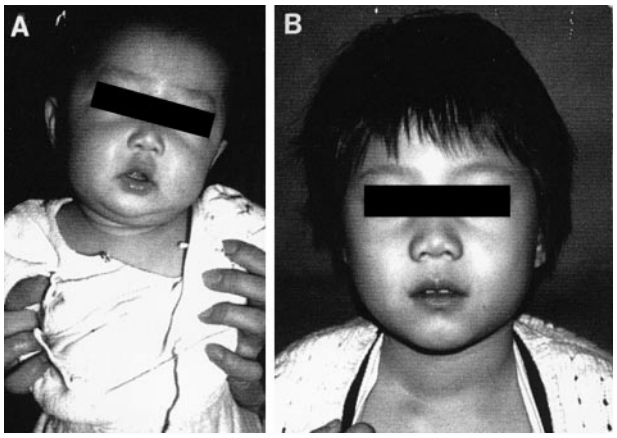

Fig 2. Sternomastoid tumor torticollis at age 3 months treated with manipulation stretching. (B) Follow-up at 4 years showing excellent improvement of craniofacial asymmetry and no residual torticollis.

the MT group, and 12.2% in the SMT group (χ^2 Exact test, $P < .001$). Multivariate analysis with the stepwise logistic regression model showed the confounding risk factors for operation as the late age at presentation group ($P < .003$), the clinical type (SMT; $P = .0124$), and the ROTGps III and IV ($P = .0006$). No cases of the POST group need surgical intervention.

DISCUSSION

The overall incidence of CMT from our previous study of over 250,000 infants was found to be 1.3%.[14] The current series differed from that of the literature in many aspects. First, we use a uniform classification system to subdivide the CMT patients into 3 clinical groups—those with SMT, the group with MT, and the POST groups. From the detailed analysis of the 1,086 cases presented early in the first year, the SMT group was the largest group (47.2%), a finding comparable with study by Canale et al[2] who reported that one third of their patients treated for CMT had a history of pseudotumor of infancy. SMT was found in this series to be associated with earlier clinical presentation, much higher incidence of breech presentation, vacuum extraction, and incidence of hip dysplasia.

Secondly, to quantitatively document the severity of the torticollis this series has used the limitation of rotation of the neck as the objective index assessment. The normal range of rotation in infants is about 110° rather than the often thought 90°. It is important to stress that the passive rotation only can be measured properly in the manner described above in the Methodology section. Such measurement was based on an interexaminer reliability study similar to that reported by Cheng and Au[14] and Binder et al.[17] In addition the study also showed that the intraclass correlation coefficient was 0.789 ($P < .001$) indicating a good correlation between rotation and passive side flexion. Hence, in this study the passive ROT was use alone for measurement of neck range in torticollis. It was clear from the results that the ROTGp IV, ie, those with limitation of over 30° had a significantly higher correlation with breech presentation, vacuum extraction rate, craniofacial asymmetry, head tilt, and earlier presentation. A total of 95.8% of the ROTGp IV were found in the SMT group in strong contrast to the POST group in which 97.5% of the cases were in the ROTGp I or II. For the hip dysplasia 0% occurred in the ROTGp I in contrast to 5.9% in the ROTGp III and 10.92% in the ROTGp IV ($P < .0001$) a finding similar to that of Binder et al[17] in 1987. In the literature, hip dysplasia was reported in 8% to 20%.[18-20] However, these papers made no further breakdown according to the severity of the torticollis. It should be noted that the presence of tumor is not always associated with a reduction in neck range. In fact, 6% of the SMT group fell into ROTGp I with no actual decrease in rotation of the neck.

The POST group in this series (22.1%) represented cases with signs of head tilt and minor or no actual deficit in the passive rotation. They resembled the group of postural torticollis as suggested by Hulbert.[6] The signs of transient head tilt probably could be secondary to abnormal fetal position without structure alteration in the sternomastoid muscle. The MT group behaves similarly to the SMT group and could represent a less severe group.

Although it is well accepted that late cases with a definite tight band of the SCM should be treated operatively, there is no clear consensus on the management of early cases. Despite the controversies, manual stretching still is the most commonly practiced treatment for SMT and MT with reported good success rates varying from 61% to 85%.[2,4,5,6,11,17,21-24] In this series, the treatments were divided into 3 groups as defined in the treatment protocol. A total of 24.5% of the CMT patients with deficits of rotation of the neck of less than 10° were treated with active home positioning program with uniformly good results with the exception of 5% of the patients who were changed to the manual stretching group with ultimate excellent to good results. A total of 75.5% (820 case) with initial limitation of passive rotation of over 10° were treated with manual stretching program in a standard protocol for a mean duration of 3.9 months with an overall final outcome of 91.1% excellent to good results. The SMT group had 12.2% fair to poor results and 7.6% necessitating surgery later. The MT group had better overall result of 6.2% poor to fair outcome and 3.1% with resultant surgery. The POST group had excellent result, and no case require surgery. Multivariate analysis also has shown clearly that the most important prognostic factors for surgical treatment were the more severe ROT group, the clinical type, and late age of presentation ($P = .0006$ to $.012$).

For patients that had significant residual limitation of rotation after manual stretching for up to 6 months, surgical treatment is recommended. In patients operated on at age 6 months to 2 years of age, excellent results can be achieved by unipolar lower pole transverse incision, excision of 1 cm of the sternomastoid tumor, careful release of all tight fascia bands, followed by an intensive postoperative physiotherapy stretching program for up to 3 months. In all the surgical cases, facial asymmetry showed significant progressive improvement with mild residual asymmetry found in less than 30% at the final assessment.[25]

The results of this study showed much better overall results with very low operation rate than most series.[2,4-6,11,15,17,21-24] This may be because of the fact that there is a clear treatment protocol and all manual stretching conducted by trained physiotherapists rather than by parents as practiced in other studies. In addition, the majority of patients in this series presented early in the first few months of life, and treatment can be started relatively early and monitored carefully in the special torticollis clinic.

Although the clinical grouping of POST, MT, and SMT may just reflect a spectrum of severity of CMT, the results of this study have shown convincingly that such grouping and assessment technique has definite clinically significant diagnostic and prognostic value and could provide a good basis for more rational study of the problem.

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