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Congenital muscular torticollis: where are we today? A retrospective analysis at a tertiary hospital

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Abstract

Background: The congenital muscular torticollis is characterized by a persistent lateral flexion of the head to the affected side and cervical rotation to the opposite side due to unilateral shortening of the sternocleidomastoid muscle. The majority of the cases resolve with conservative management, with parents/caregivers education and physical therapy.

The aim of this study was to assess demographic and clinical characteristics, treatment options, and outcome, amongst infants referred to pediatric rehabilitation consultation due to congenital muscular torticollis.

Methods: Retrospective cohort study of infants diagnosed with congenital muscular torticollis between January 2012 and December 2014. Obstetric and perinatal data, clinical presentation, comorbidities, treatment, and outcome were abstracted from clinical records.

Results: One hundred six infants were included. There was no sex predominance and mean age at first pediatric rehabilitation consultation was 11.6 (10.4) weeks. Most women were primiparous (76.4%), dystocic labor predominated (73.6%), and pelvic fetal presentation occurred in 20.8%. At examination, 49.1% of the infants had abnormalities, beyond the tilt cervical, mainly range of motion restrictions and palpable nodule in sternocleidomastoid muscle. Among the 87 children who performed the cervical ultrasound, 29 (27.4%) had anomalies. Associated clinical conditions such as hip dysplasia were identified. The majority (71.7%) were submitted to conservative treatment, 30.2% in the Pediatric Rehabilitation Department. Most infants (97.2%) showed a complete resolution of the torticollis.

Conclusions: Congenital muscular torticollis is the most common cause of torticollis in the infants. Early diagnosis, parent/caregivers education, and conservative treatment are crucial to achieving good results.

Keywords: congenital muscular torticollis, infants, physical therapy, rehabilitation

Introduction

The term *torticollis* derives from the Latin words *torquere* (twisted) and *collum* (neck) and refers to a twisted neck posture,[1] that could be acquired or congenital. The congenital form has muscular and nonmuscular causes.[2]

The congenital muscular torticollis (CMT) is the third most common congenital musculoskeletal anomaly, after dislocation of the hip and clubfoot[3,4] and refers to any deformity characterized by a persistent lateral flexion of the head to the affected side and cervical rotation to the opposite side due to unilateral shortening of the sternocleidomastoid (SCM) muscle, that is evident at birth or shortly thereafter.[4,5]

Incidence ranges from 0.3% to 2% of newborns[6,7] with a slightly male predominance (ratio of 3:2)[5] and is more common on the right side.[8] This pediatric condition may be associated with other conditions such as hip dysplasia,[9,10] plagiocephaly,[11] craniofacial asymmetry,[12] and brachial plexus injury.[13]

The etiopathogenesis of CMT is still unknown, although several theories have been proposed for its occurrence, being the compartment syndrome secondary to an intrauterine or perinatal trauma, with SCM injury that leads to ischemia and fibrosis, the most widely accepted.[14–18] Numerous obstetric and perinatal factors have been described in literature as possible risk factors for the development of CMT, such as being the first-born child, multiple gestation, breech presentation, or dystocic delivery.[14–18]

CMT is a frequent cause of referral to pediatric rehabilitation consultation. The diagnosis relies mainly on clinical and physical examination findings.[19] Further examinations may be useful to confirm a muscular origin and exclude other causes of torticollis.

The treatment of CMT is mainly conservative, consisting of parents/caregivers education (about measures of environmental modification, positioning, and home exercises) and physical therapy techniques.[7]

CMT is a frequent clinical condition but it is often under-diagnosed and referred for evaluation and treatment at later stage, compromising functional and cosmetic prognosis.

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This retrospective study aimed assesses demographic and clinical characteristics, along with treatment plan and outcome, amongst infants referred to pediatric rehabilitation consultations due to CMT.

Materials and methods

This is a retrospective cohort study of CMT infants referred to the pediatric rehabilitation consultation of a hospital in the northern region of Portugal, between January 2012 and December 2014. The study follows the "Strengthening the Reporting of Observational studies in Epidemiology (STROBE)" checklist for observational studies and reports the required information accordingly (see checklist, http://strobe-statement.org/).

Participants

The patients were eligible for the study if they were referred for evaluation at the pediatric rehabilitation consultation due to CMT and an initial sample of 115 children was assessed.

The criteria of exclusion included having other clinical conditions as cause of the torticollis, lost to follow-up, and no treatment compliance.

Data analysis

Clinical and demographic data were abstracted from clinical records: sex, mean age at first consultation, pregnancy, and neonatal data (gestational age, fetus presentation, type of labor, number of pregnancies, multiple gestation, and anthropometric measures). Side predominance of the CMT and clinical presentation at first visit including present or past abnormalities at physical examination [range of motion (ROM) restrictions, SCM tension, palpable nodule in SCM, facial asymmetry, plagiocephaly, previous history of nodule in SCM] were also collected and the presence and characterization of abnormalities at cervical ultrasound. Further information about comorbidities, type, and duration of treatment and outcome were also obtained.

Environmental adaptation measures, positioning, and home exercises were universally recommended to all parents/caregivers.

Statistical analysis was computed using SPSS for windows, version 20. We used mean (standard deviation) for descriptive analysis of normally variables. Categorical variables were presented as proportions. Associations between findings in physical examination, cervical ultrasound abnormalities, and time to resolution of CMT were studied using between-group comparisons with standard statistical testing, Chi-*square* and *T test for independent samples*, to compare proportions and means, respectively. The *P* value was set at .05.

Results

An initial sample of 115 children was assessed. Of these, 9 were excluded: 4 for having other clinical conditions that course with torticollis such as vertebral malformation, clavicle fracture, nystagmus, and cervical thymus; 4 were lost to follow-up and 1 for having abandoned treatment. So the final sample consisted of 106 infants (Fig. 1).

The mean age (standard deviation) of the 106 infants at the time of first pediatric rehabilitation consultation was 11.6 (10.4) weeks, ranging from 1 to 60 weeks after birth. There were no differences in prevalence between sex (50.9% men vs 49.1% women).

The majority was referred early after birth, mainly from neonatology consultation (43.4%) and nursery (40.6%) (Fig. 2). On the contrary, infants referred from other consultations and emergency department and from Primary Health Care were older at first consultation, ranging between 22.4 (16.2) and 31.0 (9.9) weeks, respectively.

Regarding perinatal data, fetus presentation was cephalic in the majority, with 22 (20.8%) cases having breech presentation. Labor was predominantly dystocic, 78 (73.6%), being 47 (44.3%) by cesarean and 31 (29.3%) by vacuum-assisted labor. Most women were primiparous, 81 (76.4%), with multiple gestation (gemelar pregnancy) seen in 16 (15.1%) cases.

No side predominance was seen, each side having 53 (50%) cases.

About half of CMT patients, 52 (49.1%) presented 1 or more findings in physical examination, in addition to the cervical posture alteration, more commonly passive ROM restriction in 22 (20.8%), and palpable nodule in SCM in 14 (13.2%). Regarding ultrasound, from the 87 (82.1%) infants evaluated, 29 (27.4%) showed some abnormalities. Of these, localized muscle thickening and nodule were the most common findings,

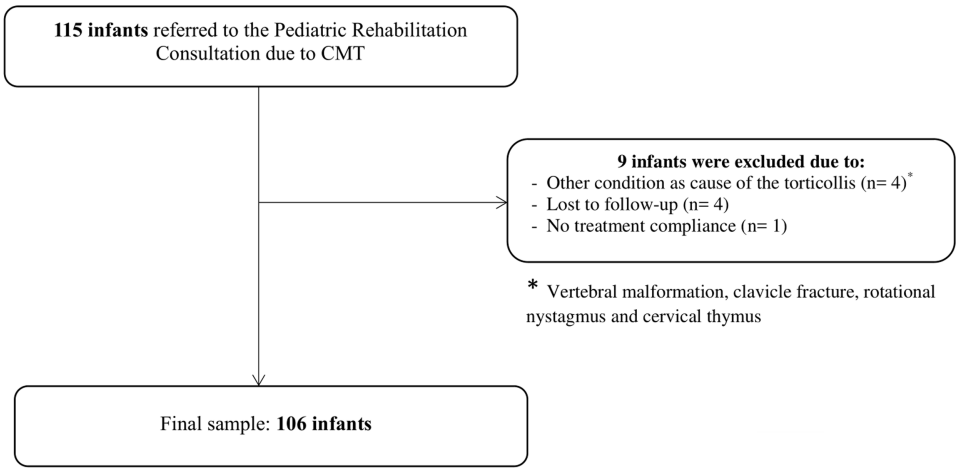

Figure 1. Flowchart of sampling procedure. CMT = congenital muscular torticollis.

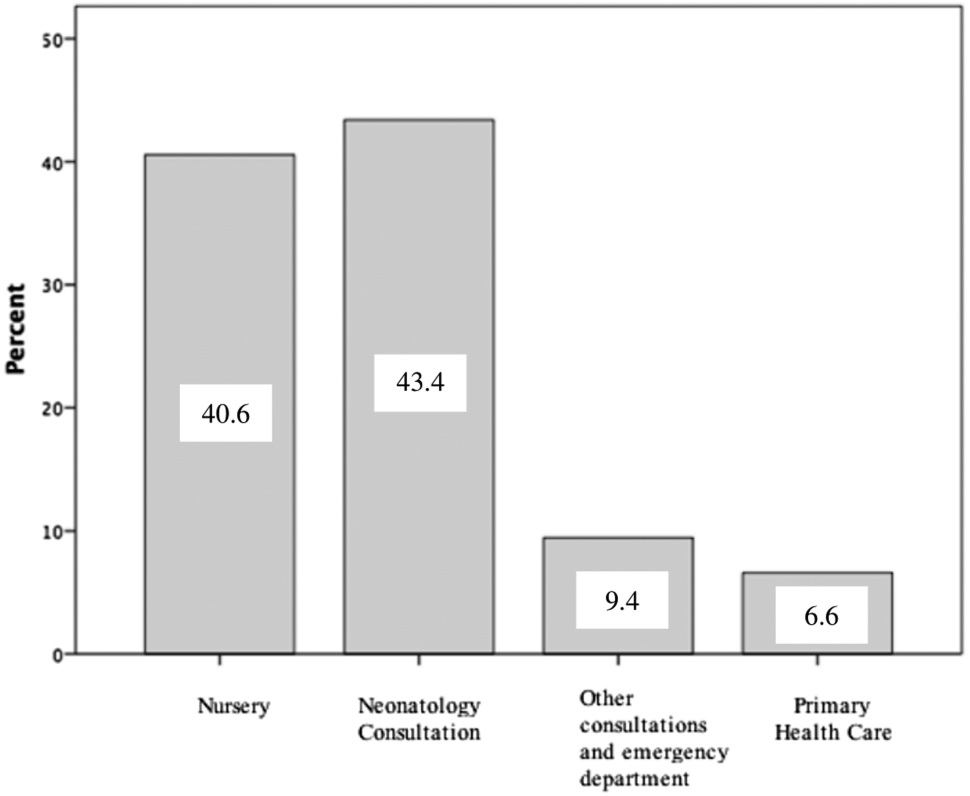

Figure 2. Referral of the infants with congenital muscular torticollis (CMT).

15 (51.7%) and 9 (31.0%), respectively, being hematoma and fibrosis other abnormalities founded at the ultrasound [4 (13.8%) and 1 (3.45%), respectively].

Additional examinations included hip ultrasound was perform in 56 (52.8%) infants with 4 (3.8%) having a Graf classification >IIa; cervical x-ray carried out in 7 (6.6%) infants who had sustained ROM restriction, all normal, and brain magnetic resonance was done in 2 (1.8%) infants, also showing normal findings.

Most infants, 98 (92.6%) had isolated CMT with the remaining 8 (7.5%) having accompanying abnormalities such as hip dysplasia [4 (3.8%)], neonatal brachial plexus palsy [2 (1.9%)], and *metatarsus adductus* [2 (1.9%)].

Regarding treatment, although all infants were submitted to environmental adaptation measures/positioning/home exercises, 76 (71.7%) had clinical indication for specific Physical and Rehabilitation Medicine (PRM) interventions, of which 32 (30.2%) were treated in the PRM Department, and the remaining did treatment at other institutions (hospital, clinical centers) (Table 1).

Table 1

Proportion of infants submitted to physical and rehabilitation medicine treatment by treatment setting

Type of treatment	N (%)
No treatment*	30 (28.3%)
Treatment setting (PRM treatment)	76 (71.7%)
PRM department	32 (30.2%)
Other institution	44 (41.5%)

PRM = physical and rehabilitation medicine.
* All the infants underwent environmental adaptations measures/positioning/home exercises.

Infants initiated PRM treatment at mean age of 14.3 (10.0) weeks, earlier for those referred from nursery [8.0 (4.92) weeks] and later for referrals from primary care physician or assisting pediatrician, with a mean age of 35.3 (8.9) weeks.

The mean time to resolution of the CMT was similar in the children that made treatment in PRM Department or in other institution, 19.9 (8.77) and 18.7 (10.4) weeks, respectively, and most of the children had a complete resolution 103 (97.2%) (Fig. 3). None of the studied infants required surgical intervention.

Mean time to resolution of CMT was higher for those with abnormalities at physical examination compared to those without findings other than cervical tilt at physical examination [21.5 (10.6) vs 16.2 (7.6) weeks, $P = .02$].

In the group of the 76 children who underwent PRM treatment, presence of abnormalities on cervical ultrasound was not associated with longer treatment duration [19.0 (9.2) vs 20.5 (10.2) weeks, $P = .55$)] (Table 2).

A positive association was found between findings in physical examination and ultrasound abnormalities with 14 (48.3%) patients having both physical examination and ultrasound changes involving the SCM ($P < .05$), whereas 15 (51.7%) of those with normal physical examination had an abnormal cervical ultrasound (Table 3). Detection of an abnormal SCM at physical examination had a positive predictive value of 60.9% and negative predictive value of 76.6%.

Discussion

CMT is the most common form of torticollis in infants and could be classified as 1 of 3 types, according the findings at physical examination and in order of increasing severity as postural CMT,

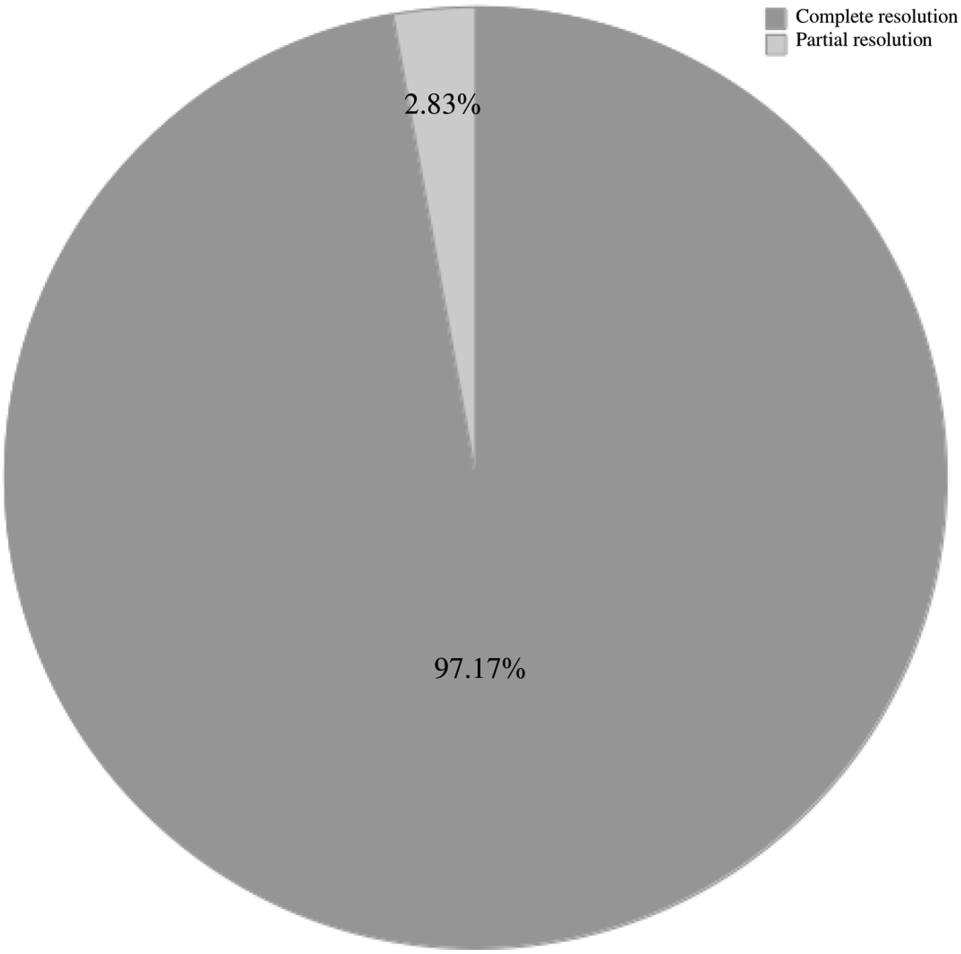

Figure 3. Resolution of congenital muscular torticollis (CMT).

the mildest form[6,20,21]; muscular CMT and SCM mass CMT, the most severe presentation.[6,20,21]

With this retrospective analysis we have shown that CMT remains an important reason for referral to PRM, especially in the presence of breech presentation, primiparity, dystocic labor, and multiple gestation. Significant associations were found between findings in physical examination, abnormalities in cervical ultrasound, and time to resolution.

We found slight male predominance but, despite reported right-side preference in other studies,[5] we found no side preference of the CMT.

Despite being a studied entity, its etiopathogenesis is still unknown. The development of CMT was, however, found in several series as being associated with numerous obstetric and perinatal factors.[14–18] Higher prevalence of breech presentation is reported in children with CMT than in general population and this finding was seen in 20.8% of the 106 children with CMT. Other risk factors described in literature are multiple pregnancy, observed in 15.1% infants of the study, primiparity, detected in 76.4% and dystocic labor, evident in 73.6% of the infants.

Many reports refer an association between CMT and other clinical entities. The coexistence of hip dysplasia and CMT is described in literature, ranging between 0% and 29%.[9] Amongst the 106 children, 3.8% had concomitant hip dysplasia. This association and the clinical and functional impact of hip dysplasia warrant systematic screening of this entity in children with CMT.

In this study, it was found that presence of abnormalities at initial physical examination was more frequently associated with

Table 2

Abnormalities at cervical ultrasound and time to resolution of the congenital muscular torticollis

	Cervical ultrasound	N	Mean	Std. deviation	*P*
Time to resolution (wk)	With abnormalities*	26	19.00	9.239	.55
	Normal	43	20.49	10.229	

CMT = congenital muscular torticollis.
* Thickening, nodule, hematoma, or fibrosis of the sternocleidomastoid muscle.

Table 3

Abnormalities at shortening of the sternocleidomastoid (at physical examination) and abnormalities at cervical ultrasound

	Cervical ultrasound findings†		
SCM findings*	Normal (n = 58)	With abnormalities (n = 29)	*P*
Normal	49 (76.6%)	15 (51.7%)	.001
With abnormalities	9 (39.1%)	14 (48.3%)	

SCM = sternocleidomastoid muscle.
* Muscle tension or nodule.
† Thickening, nodule, hematoma, or fibrosis of the sternocleidomastoid muscle.

abnormal cervical ultrasound and longer time to resolution of CMT compared to those only having cervical tilt. Therefore, physical examination is an essential diagnosis measure of CMT and could be a good predictor of the prognosis of CMT.

So, the above correlations may indicate that (1) decisions guiding treatment are based on clinical examination rather than on ultrasound findings, (2) presence of ultrasound abnormalities frequently lack prognostic significance in predicting CMT clinical recovery.

The treatment of CMT consists, namely, in PRM measures that aims, initially, in environmental modification measures, positioning, and home exercises carried out by the parents/caregivers.[6] Physical therapy interventions include postural correction, passive and active stretching of the tight SCM, optimization of cervical active, and passive ROM and strengthening of weak neck and trunk muscles.[7]

In general, infants with postural CMT, identified in an early phase, usually have a rapid and complete resolution of torticollis. On the contrary, those who are diagnosed later, after 3 to 6 months of age and those having the most severe form of CMT (SCM mass) usually have the longest period of PRM treatment.[5,22,23] However, in the retrospective analysis performed, these correlations were not always evident. Some children with an early intervention and minor abnormalities at clinical examination (cervical tilt) had longer periods of treatment than children with major abnormalities at physical examination or in cervical ultrasound. This kind of findings can be explained by the limitations founded in the study. First, the heterogeneity of the population, although it gives a clearer picture of CMT in clinical practice it affects interpretation of results and make it harder to detect differences. Secondly, the different treatment settings, where treatment outside hospital settings was more difficult to study due to lack of reliable information.

Conclusions

CMT is a frequent condition in the infants at birth or soon after. Early diagnosis and interventions with appropriate treatment and follow-up is critical for early correction and prevention of future complications.

Despite the torticollis being a condition with a good prognosis, its management can be difficult and is not always linear. A careful medical assessment is essential to exclude other causes of torticollis as CMT may be a warning sign of underlying conditions that require urgent interventions.

Given the limitations acknowledged above, further studies with a more homogeneous population should be conducted to provide a more representative and statistical analysis.

Conflicts of interest

The authors report no conflicts of interest.

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