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Annals of Physiotherapy & Occupational Therapy
ISSN: 2640-2734

Congenital Muscular Torticollis-Long-Term Consequences

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Research Article

Volume 4 Issue 2

Received Date: March 18, 2021

Published Date: April 01, 2021

DOI: 10.23880/aphot-16000193

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Abstract

Background: Congenital muscular torticollis is the most common type of congenital torticollis. A good prognosis is expected, especially if treated early. However the occurrence of musculoskeletal and psychomotor development sequelae, in the medium to long term, has been reported in the literature.

Purpose: To review the current scientific knowledge on the subject and to characterize the late term consequences of congenital muscular torticollis in the population followed in a paediatric tertiary Hospital.

Methods: A bibliographic research was carried out and it was performed a review of the electronic records of the patients evaluated in 2009. Additionally, a telephone questionnaire was conducted, aiming to characterize musculoskeletal and developmental changes occurring during the last eleven years.

Findings: There is a scarcity of studies addressing congenital muscular torticollis sequelae in the medium/long term, and the studies present very low levels of evidence. Sequelae describedare in the musculoskeletal, visuospatial, cognitive-behavioral and psychomotor spectrum. We identified 41 patients with congenital muscular torticollis newly diagnosed in our outpatient clinic in 2009, of which 26 answered the telephone query. Age at the time of the query ranged from 11,5 and 14 years. At the present time, 65% of patients had visual and/or dentition/chewing changes.

Conclusions: In the series of patients studied, vision and oromandibular problems, followed by neurodevelopmental changes, were the most reported by the parents. The bibliographic references converge in the need for periodic reassessments in children with severe / difficult to resolve congenital muscular torticollis or severe skull deformities, in order to detect long term spinal and psychomotor development sequelae.

Keywords: Congenital Muscular Torticollis; Sequelae; Development; Cognitive Impairment; Scoliosis

Introduction

Congenital torticollis (CT) can be caused by several pathological entities. A fifth of CT is non- muscular, justifying a careful clinical assessment of the child before starting treatment and regular medical follow-up [1-3].

Congenital muscular torticollis (CMT) is, within the possible differential diagnoses, the most common type of congenital torticollis, with an incidence of 0.3-3.92% and a male: female ratio of 3:2.4.4 This is the third most frequent congenital musculoskeletal disorder, after hip developmental dysplasia and metatarsus adutus/clubfoot. Although the pathophysiology is not fully understood, clinically there is an injury to one of the sternocleidomastoid muscles (SCM), with ipsilateral inclination and contralateral rotation of the head [4,5].

The diagnosis is clinical, with complementary diagnostic tests being exceptionally applied to exclude secondary causes of CT.

It is believed that CMT is pathology with a good prognosis if treated early. First line of treatment is conservative and

consists in educating the caregivers regarding environmental changes, child positioning and home exercises, with or without physical therapy [3,4].

Usually, children with early identification of CMT have a quick and complete resolution. Late diagnosis, meaning after six months of age, cervical movements asymmetry superior to 15º or presence of SCM nodule, are generally associated with longer treatment periods and worse prognosis [5-8].

The most frequent short-term sequelae are plagiocephaly. This deformity frequently occurs in association with CMT, due to the preferably unilateral pressure on the immature skull of the newborn [9,10].

It presents as a parietoccipital flattening contralateral to the CMT, with or without frontal protrusion, anteriorization of the pinna and possibly hypometry of the face (eye and jaw), all on the same side of the flattening [11,12].

There has been an increased recognition of medium/ long term sequelae, namely scoliosis [13-15] in the musculoskeletal (MSK) spectrum and visuospatial, cognitive-behavioral psychomotor alterations in the development spectrum [16-19] and While MSK sequelae are mostly explained by residual asymmetry, psychomotor development consequences seem to also affect children with favorable clinical evolutions, meaning symmetry of the active and passive cervical rotation and inclination, completely or with a difference of less than 10º [7,20].

From a non-MSK or developmental perspective, global sensorimotor development and, more specifically, the child's ability to acquire postural symmetry should be taken into account. This is the necessary basis for the development of postural and movement references, that is, only through symmetry will the child be able to acquire reactions of balance and ability to transfer body weight, among others, which will allow him to reach progressively more complex stages in perceptual and motor field [16].

Our main goals are to revise the current knowledge on medium/long term sequelae of CMT described in literature and to study their prevalence in the population followed in a pediatric tertiary hospital. This knowledge is relevant for assessing prognosis and deciding the most adequate follow-up of these patients.

Material and Methods

A bibliographic research was performed in the Google Scholar, Pubmed, and Cochrane Database of Reviews and PEDro databases, using the keywords described above.

The search carried out on Google Scholar was restricted to the period from 2018 to the present date. Studies investigating the occurrence of sequelae determined by the CMT, both in pediatric and adult age, were selected.

We excluded studies whose subject was only the assessment and treatment of CMT without mention of possible sequelae in the medium / long term, as well as articles on sequelae of non- muscular CT.

A cross-sectional study was performed by telephone inquiry. The patients were selected using the informatic records from our hospital. Inclusion criteria were: being observed for the first time by a Physical Medicine and Rehabilitation (PM&R) doctor in 2009, due to CMT diagnosis. All cases of non-muscular torticollis and prematurity were excluded. The inquiry was performed by telephone, to one of the parents, characterizing aesthetic, spinal alignment, psycho-motor and cognitive-behavioral changes that occurred until September 2020, representing a period of approximately 11 years after the diagnosis. A descriptive analysis of the results is presented.

Results

The bibliographic research returned the following articles according to the keywords / key phrases used, as shown in Table 1.

Key words/ phrases	Database	References total	References selected by relevance
"congenital torticollis" and "sequelae"	PubMed	49	10
	Cochrane Database of Reviews	1	0
	PEDro	0	0
	Scholar Google **	38	4
"congenital torticollis" and "child development"	PubMed	11	5
	Cochrane Database of Reviews	0	0
	PEDro	0	0
	Scholar Google **	222	11

"congenital torticollis" and "scoliosis"	PubMed	8	4
	Cochrane Database of Reviews	0	0
	PEDro	0	0
	Scholar Google **	94	6
"congenital torticollis" and "cognitive impairment"	PubMed	0	0
	Cochrane Database of Reviews	0	0
	PEDro	0	0
	Scholar Google**	42	3
			Sub-TOTAL: 43 Duplicate references: 15 TOTAL: 28

** Research conducted in November 2020. Restricted to the 2018-2020 interval, and searching for key words/phrases in any area of the text.

Table 1: Bibliographic research articles according to the keywords/key phrase and data are used.

The twenty-eight selected articles are distributed, regarding the type of publication, as shown in Table 2.

CMT and...	ditorials/Opinion/ Co mmentar/ Perspectives	Textbook chapter	Case Reports and Series	Retrospective Cohort study	Review Article	Cohort Study (Prospective Observational Study)	Randomized Controlled Trial	Systematic Review	Meta-Analysis
Sequelae	(8) Sargent B, 2019 - Am Acad Pediatrics	(10) Massimi Textbook of pediatric Neurosurgery, 2020 - Springer (4) Philips J- ... of Disorders of the Child's ..., 2018 - Springer (40) Eranhikkal A - International Journal of Clinical..., 2020	(13) Hussein MA, Yun IS, Park H, Kim YO. J Craniofac Surg. 2017 Jan;28(1):46-50. (39) Tonkaboni, B Mirzashahi Orthopaedic Surgery and ..., 2018 - researchgate.net (41) Ferguson JW. Int J Oral Maxillofac Surg. 1993 Feb;22(1):7-10	(35) Chate RA. Br J Oral Maxillofac Surg. 2005 Oct;43(5): 428-34 (43) Ahn AR, et al J Craniofac Surg. 2018 May;29(3):e327-e331 (32) Kim HG, Yim SY. Cleft Palate Craniofac J. 2019 Nov;56(10):1295-1301. (33) Seo SJ, et al Plast Reconstr Surg. 2013 Aug;132(2):407-13. (24) Binder H, Eng GD, Gaiser JF, Koch B.Arch Phys Med Rehabil. 1987 Apr;68(4):222-5. (38) Lim KS, Shim JS, Lee YS. Clin Orthop Relat Res. 2014 Apr;472(4):1271- 8. (3) DM Amaral, RPBS Cadilha, JAGM Rocha... -Porto biomedical ..., 2019 -ncbi.nlm.nih.gov	Gundrathi J, Cunha B, Mendez MD. Congenital Torticollis. [Updated 2020 Nov 27]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2020 Jan-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK549778/ White KK, Bouchard M, Goldberg MJ. Common ... Avery's Diseases of the Newborn. 10th ed. Philadelphia, PA: Elsevier; 2018:chap 101.	(34) Lee JK, et al J Bone Joint Surg Am. 2012 Jul 3;94(13):e93 (42) Vallen, T Xi, M Nienhuijs, W Borstlap... - International Journal of ..., 2020 - Elsevier.		(30) Kuo AA, Tritasavit S, Graham JM, Jr.Pediatr Rev. 2014 Feb;35(2):79-87	
Child development/ Cognitive impairment			(28) Schertz M, Zuk L, Green D. J Child Neurol. 2013Oct;28(10):1215-21.	(25) Watemberg N, Ben-Sasson A, Goldfarb R. Pediatr Neurol. 2016 Jun;59:36-40. (9) MA Hussein, TWoo, IS Yun, H Park, YO Kim -Journal of Plastic..., 2018		(27) Öhman A, Beckung E. PM R. 2013Oct;5(10):850-5. (17) BR Collett, D Kartin, ER Wallace... -Pediatric Physical ..., 2020 -journals.lww.com		(16) Tessmer A,Mooney P, Pelland L. Pediatr Phys Ther. 2010 Winter; 22(4):378-83.	
Scoliosis		(14) MA Hussein, IS Yun, D won Lee, H Park... - Journal of Craniofacial ...,2018	Hobaek Siegenthaler M. J Chiropr Med. 2017 Sep;16(3):257-261.	(43) Kim JH, Yum TH, Shim JS. Clin Orthop Surg. 2019 Sep;11(3):344-351					

Table 2: Distribution of the selected articles according to type of publication (abbreviated bibliographic references, numbered as showing in References section).

1. The shape of your child's head has worried you for the past year?	YES 2/26
2. Do you notice any cranial deformity?	YES 2/26
3. Do you notice limited neck movements or a preferred position?	YES 4/26
4. Did your child have complaints of head / neck / spine pain?	YES 7/26
5. Has your child ever been a victim of bullying due to the shape / size / position of his/her head?	NO 26/26

Table 3: Present signs/symptoms.

6. The family doctor / pediatrician was ever concerned with the development?		YES 3/26
7. Before entering primary school did your child already: Ride a tricycle? Make drawings? Say age / address / name	Dress alone?	NO 2/26
	Used the bathroom by himself?	
	Eat by himself?	
8. Does your child jump and run as quickly and easily as boys of the same age?		NO 3/26
9. Does your child write / draw fast enough to accompany the other children in the classroom?		NO 5/26

Table 4: Child development.

10. Does your child have interest in and enjoy participating in sports activities that require good motor coordination? Does he practice regularly? 15/26*	YES 23/26*
11. Does your child play any musical instrument?	YES 4/26
12. Does your child have good school performance? Failed? YES 4/26** Has school support? YES 5/26**	NO 6/26**

Table 5: Activities / Participation.

13. Scoliosis?		YES 0/26
14. Chewing/ jaw pain?		YES 2/26
15. Grinding teeth issues?		YES 5/26
16. Dental issues?		YES 10/26
17. Vision issues?		YES 12/26
18. Earing issues?		YES 4/26
19. Septal / snoring issues ?		YES 9/26
20. Active problems:	Past problems:	YES 13/26
● **Orthopedic:** ● Postural scoliosis ● Short stature ● Flat foot	● Cardiac remite ● Speech disorder ● ENT surgery	
● **Developmental**: ● Autism ● Dyslexia		
● **ENT/ Dentistry:** ● Neurossensorial hypoaccusia ● Prognathism ● Cross bite ● Traumatic mandibulary joint dysfunction		
● **Other:** ● Asthma ● Precocious puberty ● Hypospadia ● Oftalmological migraine		
21. Maintains follow-up appointments in medical specialities?		YES 12/26

ENT- Ears, Nose and Throat.

Table 6: Medical problems diagnosed after PM&R discharge.

A total of 41 children, with term pregnancies, were observed in a first PM&R consultation during 2009 due to CMT. From these, we were able to reach 28 parents, and all agree to answer the inquiry (Tables 3-6). One child was excluded because of a posterior diagnosis of ocular torticollis and one child was excluded for a history of prematurity that was not mentioned on our informatic records.

For descriptive analyses, 26 inquiries were considered, regarding 13 males and 13 females, with an average age of 11.5 years at the time of the inquiry. The first observation by the PM&R Doctor was done at three months of age (minimum 12 days, maximum 17 months), with a mode of one month and median of two months.

They had no mention of relevant personal history. Twelve presented plagiocephaly at the time of the first observation, two did not, and in 12 there was no mention of cranial symmetry in the first observation.

All patients were integrated into home treatments performed by the parents, together or without physiotherapy in the PM&R Department. In one of the patients, surgical treatment with unipolar tenotomy was required at nine years of age.

Follow-up was typically maintained until a difference of less than 10º, both in active and passive cervical inclination and rotation, together with an age-appropriate psychomotor development was achieved.

In our sample the mean age at discharge was 15 months (minimum three months and maximum nine years), with mode and median of 10 months. All patients had complete or acceptable symmetry of the skull and normal psychomotor development according to the Mary Sheridan scale, except for a boy with delayed psychomotor development. He was referred for Development consultation and posteriorly diagnosed with autism.

At the time of the inquiry, six parents (23%) reported that their child presents changes in global motor development, manual dexterity, and/or difficulty following classes/ poor school performance. Of those, four had plagiocephaly described on the first observation and two had no record of cranial symmetry.

Seventeen patients (65%) had changes in vision, dentition or chewing. In the subgroup of patients with CMT-associated plagiocephaly (n = 12), nine were found to have at least one of these manifestations. Four had isolated oromandibular changes (prognathism, cross bite, dental crowding and bruxism) and three isolated visual changes (use of visual correction with glasses, denying asymmetry in the adaptation of the glasses to the face) and two manifested changes in both oromandibular and visual changes. The two patients where plagiocephaly was denied, none had oromandibular problems and one has vision changes. In the subgroup of 12 patients with no record of cranial symmetry, seven had at least one of these manifestations: two had isolated oromandibular changes (crowding), one had isolated visual changes and four manifested changes in both sectors.

None of the 26 patients was diagnosed with scoliosis until now. Parents described a diversity of other problems, listed on Table 6.

Sports practice was frequente with 15 patients (57%) practicing sports of medium to high difficulty.

Discussion

Back to sleep policy, in prevention of sudden death, exponentially increased postural deformities of the skull, with up to 20 to 30% of children affected in the first six months of life [9]. Children with cervical alignment deviations, with or without positional deformities of the skull, should be seen by PM&R during the first three months of life, in order to be integrated into appropriate treatments at an early stage [5-7]. In our study, the date of the first observation was within the current guidelines, reflecting that both primary care and pediatric doctors are alert to head deformities and abnormal cervical positioning in the first months of life.

Plagiocephaly association with CMT is well described in the literature. In our series, at least half of the patients presented plagiocephaly associated with CMT, although the exact number could not be found because medical records had limited information. This also limits our understanding of the progression of this patient's plagiocephaly, since the evaluation of cranial deformities should be performed in a reproducible way. An observational classification should be applied, using the Argenta scale, or measuring with a craniometer and comparing the cephalometric indices with a graph adjusted to the studied population. This serial comparison of the values obtained would allow a more concrete assessment of the evolution of the deformity [7].

Being CMT such a frequent pathology, we found a relatively low number of publications on sequelae in the medium / long term on the literature, and with very low levels of evidence (Levels 3 to 5 according to the Oxford Center for Evidence Based Medicine). This may be explained by a low frequency of medium / long term sequelae, but can also be the case that non-immediate sequelae are being neglected or not being associated with the history of CMT in the early childhood.

Although CMT and plagiocephaly normally have a spontaneous resolution within the first two years of life, their occurrence may be associated with changes in the child's neurodevelopment even after deformity resolution [21-24], although this direct relationship has not always been found [7].

Watemberg, et al. [25] in their retrospective study of 173 CMT cases, found that a quarter had motor asymmetry and plagiocephaly, with a slight delay in global motor acquisitions being more common. Of these children, 82% showed no motor asymmetry at two years of age, and for that reason, the asymmetry was considered to be benign and transitory.

Ohman, et al. [19] concluded, in their prospective study with 122 children between two and 18 month of age, that CMT is a risk factor for delayed motor development, but only up to 10 months of age [26]. These researchers followed the same group of children, and assessed them at preschool age (between three and a half, and five years) and did not identify changes in the development of these children [27].

In contrast, Collett, et al. [17] in a prospective study of school-age children, found that those who had a moderate to severe cranial deformity in early childhood, maintained global motor and manual dexterity /hand coordination asymmetries at entrance to primary school, and that vigilance in the most determinant school stages is advised. Also Schertz, in his long-term assessment of the neurodevelopment of children with CMT, found that 44% of 68 patients, with ages between seven and nine years, had or were at risk of developing a neurodevelopmental disorder such as hyperactivity and attention deficit, changes in coordination, language or disturbance in the autism spectrum. In this study, half of the children were accessed by the authors and the other half was only submitted to a telephone interview. The percentage of patients identified as having neurodevelopmental disorders in the telephone interview was much lower, alerting about the inability of parents / caregivers to perceive slight changes in children [28].

In the 2014 systematic review article by Kuo AA, et al. [29] the authors conclude that there is grade C evidence, based on observational studies, for active CMT exploration whenever there are pre and postnatal risk factors, such as intrauterine malposition, oligoamnios and instrumented delivery. They also report the same degree of evidence for the association of sequelae associated with CMT, such as plagiocephaly and global motor delay, and for the need for prone decubitus moments. They found grade D evidence (based on expert consensus) for early referral to a physiotherapist experienced in the pathology.

Permanent deformities of the skull and face may occur and result in aesthetic and / or functional changes, with greater or lesser severity according to the degree of asymmetry. Chung-Chih Yu, et al. and colleagues, in their study of 14 patients with untreated CMT (aged between one month and 24 years), found that changes in the skull cap, bones at the base of the skull and bones in the face, start during the first year and affect more the posterior fossa of the skull, and that asymmetries of the facial bones at five years of age tend to worsen throughout life, particularly during adolescence [30]. Similar conclusions are mentioned in other studies [31,32].

Lee, et al. [33] prospectively studied, in 2012, a cohort of 80 CMT patients after release of the SCM. They found an improvement in craniofacial deformity that was more evident in the first postoperative year than in the second and mainly in the group under the age of five years.33 Other authors describe good aesthetic and functional results after SCM release in patients up to 10 years of age.

Several other authors describe cranio-maxillary asymmetries/deformities, namely lateral mandibular deviation, dysplasia of the mandibular fossa of the temporal bone, shortening of the mandibular condyle neck, irregular angulation of the mandibular condyle, dental malocclusion, lateral crossbite and spasm of masticatory muscles [15,35-39].

The best way for a correct and reproducible measurement of the skull and face measurements and their evolution after treatment has also been the subject of studies, as the ones by Ferguson, et al. [40,41]. Regarding the telephone questionnaire performed, it integrated questions regarding cranial and facial residual asymmetry.

A significant percentage of patients (65%) presented, in pre-adolescent age, changes that may be related to craniofacial asymmetries, in agreement with the studies mentioned above. The possibility of reassessing these patients in this regard would be interesting to complement the present study.

The small sample size, along with the fact that in about half of the cases there is no record that confirms or denies skull deformities at the time of CMT diagnosis, does not allow us to perceive a relationship between cranial / facial asymmetry and the manifestations mentioned above.

The occurrence of spinal postural or static deviations in the medium to long term in association to CMT, is a concern often manifested not only by parents, but often by health professionals.

Hussein and collaborators, in 2018, studied a group

of 15 patients under the age of eight years, evaluating the cervical spine in 3D computed tomography and comparing anatomical and volumetric asymmetries of the vertebrae in each spacial plane. They concluded that, in children with untreated CMT, vertebral bone asymmetries begin toestablish as early as eight months of age, with the axis being the first vertebra to be affected. They also state that the severity of vertebral deformity is directly related to age and to the degree of shortening of the SCM [14].

The same author described a group of four adults with a history of untreated CMT, revealing anatomical changes in the atlas and axis, namely in terms of the articular facets and volumetry of the sagittal hemibodies of these vertebrae. Similar changes, but progressively minor, were found up to the sixth cervical vertebra. These changes determined joint limitations in cervical rotation and inclination [13].

Ahn AR, et al. [42] in their retrospective study of a cohort of 41 patients with SCM tenotomy at the mean age of eight years, found atlanto-axial rotation in 82.76% of the cases.

Kim JH, et al. [43] in a retrospective study of 87 patients, analyzed the development of cervicothoracic scoliosis in patients with CMT and its evolution after the release of SCM. The patients were between five and 44 years old. He reports an 82% prevalence of cases of cervicothoracic scoliosis in his study group, having found a statistically significant improvement, after surgery, in the cervicomandibular and cervicothoracic angle, in the group of patients submitted to surgery under the age of 15 years.

In our series, vision and oromandibular problems, followed by the neurodevelopmental changes, were the most reported by the parents.

From our bibliographic research, we can conclude that CMT, despite being so common in Pediatric Rehabilitation, lacks further study of its sequelae in the medium to long term. Although scarce, the bibliography found seems to converge to the need for periodic reevaluations in two subgroups of patients: those with severe / difficult to resolve CMT, and those with severe skull deformities.

Each subgroup of patients is likely to benefit, respectively, from evaluations of spinal static alignment and psychomotor development. The first, in pre-adolescence with regular assessments during the rapid growth phase of adolescence, and the second in the pre-school phase, with reevaluations at least at each educational cycle.

More studies are needed to understand the long-term evolution of these children, requiring series with a larger number of patients and followed for a longer period of time.

Acknowledgement

The authors would like to thank Dr. Ana Soudo for all the availability on reviewing the manuscript.

Statement of Conflict of Interests

The authors have no conflict of interests.

Funding

Nothing to declare.

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