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Research Article

Dentofacial Complications of Congenital Muscular Torticollis in Children Up to 5 Years of Age

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Abstract

Introduction: Dentofacial complications of congenital muscular torticollis can affect daily and fundamental activities such as chewing and phonation.

Objective: To characterize the dentofacial complications of congenital muscular torticollis and determine the ages at which they transit.

Methods: A descriptive, cross-sectional, and retrospective study was conducted from January 2017 to December 2020. The universe consisted of 220 patients under 5 years of age, with a diagnosis of congenital muscular torticollis, followed in the provincial consultation of Neurodevelopment of the South Children's Hospital "Antonio María Béguez César" in Santiago de Cuba, Cuba; and the sample was made up of 162 patients followed for a minimum of one year, selected probabilistically by simple random sampling.

Results: Dentofacial complications occurred in 20.98 % of patients. Men predominated in both complicated and uncomplicated patients (58.8 and 60.2 %) respectively. Plagiocephaly (12.3%) and facial asymmetry (11.7%) were the most frequent sequelae. There was a statistically significant association of complications with the increase in the mean age ($p < 0.01$) and the median age of the patients ($p < 0.03$). Plagiocephaly was the earliest onset sequelae with a mean age of 6.6 months (SD = 0.5), while atrial dystopia was the latest with a mean of 11.1 months (SD = 1.8).

Conclusions: The increase in the average age at diagnosis and treatment of congenital muscular torticollis increases the risk of dentofacial complications.

Keywords: Torticollis; Complications; Dentofacial

Introduction

Congenital muscular torticollis is a postural deformity that is manifested by ipsilateral cervical lateral flexion and contralateral cervical rotation due to unilateral shortening of the sternocleidomastoid muscle [1].

The disease burden of congenital muscular torticollis is high, being considered the third most commonly diagnosed musculoskeletal anomaly in childhood, with an incidence in newborns ranging from 0.3 to 2%; and its current trend is to increase, it also represents 81.6% of all confirmed torticollis in children [1-4].

Early diagnosis and rehabilitation treatment with massages and stretching exercises manage to avoid complications of the disease in up to 90% of patients, the remaining 10% require surgery.[4] It is recommended to start physiotherapy treatment before three months of age, although ideally in the first month of life [1,4].

If congenital muscular torticollis is not detected at an early age and, therefore, is not treated early, it is common for its dentofacial organic complications to become evident during growth, including: craniofacial asymmetries, plagiocephalic frontal deformation (greater growth of the skull on the opposite side of torticollis), orbital dystopia (the eye on the unaffected side is higher), atrial dystopia or posterior displacement of the ear on the affected side and malocclusion (anomaly in the alignment of the teeth and dental arches that can affect chewing function and dental aesthetics) [3,5,6].

These dentofacial deformities can limit, and in many cases condition, in a decisive way, habitual and fundamental activities of daily life such as phonation and chewing, in addition to generating pain and functional disability of the temporomandibular joint [5].

The Maternal and Child Care Program is prioritized by the Cuban Health System. Considering the above and the importance of early evaluation and intervention through the identification of risk factors, in the province of Santiago de Cuba, Cuba, there are no recent studies on the behavior of dentofacial complications of congenital muscular torticollis in children. The objective was to characterize the dentofacial complications of congenital muscular torticollis and determine the ages at which they transit.

Methods

A descriptive, cross-sectional and retrospective study was carried out in patients from one month to 5 years of age diagnosed with congenital muscular torticollis, attended at the provincial Neurodevelopment clinic of the South Children's Hospital "Antonio María Béguez César" in Santiago de Cuba, Cuba, in the period from January 2017 to December 2020.

The universe or eligible population consisted of the 220 children diagnosed with congenital muscular torticollis seen at the provincial Neurodevelopmental Clinic in the aforementioned period. The minimum sample size was calculated using the EPIDAT version 4.2 statistical package, and was made up of 162 patients who were selected through a simple random sampling.

Inclusion Criteria

- Children up to 5 years of age, who had a minimum observation period of one year after their diagnosis and whose clinical record included all the variables to be investigated.

Exclusion Criteria

- Having an associated disease, other than congenital muscular torticollis, as a cause of dentofacial complications.

Data collection was carried out by reviewing the individual medical records of the outpatient clinic. A form was prepared for data collection with the variables under study, which were taken directly from the medical records in the Archive Department of the South Children's Hospital, by the main author of the research.

The dichotomous nominal qualitative variables were: sex (female, male) and evolution (with complications, no complications). The nominal polytomic qualitative variable was: presence of complication (one or more of the following were taken into account: plagiocephaly, facial asymmetry, ocular dystopia, atrial dystopia and malocclusion). The age at diagnosis and initiation of treatment of the disease was considered discrete quantitative (the age reached in months or years was used).

The data obtained were entered into a database in Microsoft Access (Microsoft Corporation, Washington, United States). It was used in the processing of Microsoft Excel and the EPIDAT statistical package (Program for the Analysis of Tabulated Data), version 4.2. The qualitative variables were expressed in whole numbers and percentages and the quantitative variables in median, mean and their standard deviation, estimates were made by interval at 95 % for the arithmetic mean. Student's t-test was also applied to compare the means and establish if there were significant differences between the average ages of the patients, to determine the associations between variables, Fisher's exact test *and* Pearson's *chi-square* test were used, in all cases the significance level (α) of 0.05 was used.

It is a retrospective study so the use of parental informed consent was not necessary. The authors declare their commitment to confidentiality and protection of the information collected during the research. Authorization was also requested from the management of the center and the approval of the Research Ethics Committee and the Scientific Council for the execution of the study.

Results

Of a total of 162 patients analyzed in the study period, 34 presented dentofacial complications of congenital muscular torticollis (20.98%).

Male sex predominated in the study in both patients with dentofacial complications (58.8%) and those who did not (60.2%). There was no statistically significant association of complications with sex (Table 1).

Sex	Patients				p*
	With complications (n = 34)		No complications (n = 128)		
	No	%	No	%	
Male	20	58,8	77	60,2	0,575
Female	14	41,2	51	39,8	

Table 1: Patients with and without complications by sex.

Legend:

*Fisher's exact test.

% Calculated based on the total of the columns.

Source: Medical records.

The percentage distribution of dentofacial complications of congenital muscular torticollis was shown in Table 2, with plagiocephaly and facial asymmetry being the most frequent, in 12.3 and 11.7 %, respectively, of the total sample.

Of the 34 complicated patients, 14 presented two or more dentofacial complications of the disease simultaneously for 41.2% of the total complications, the associations with the highest observance were plagiocephaly with asymmetry and asymmetry with ocular dystopia.

Table 3 shows the mean age of patients with complications (7.62 ± 2 months), which exceeded the overall age (6.57 ± 2 months) and that of patients without complications (5.52 ± 2

Complications	Patients	
	No	%
Plagiocephaly	20	12,3
Facial asymmetry	19	11,7
Eye dystopia	10	6,2
Distopia auricular	9	5,5
Malocclusion	1	0,6

Table 2: Percentage distribution of dentofacial complications.

Legend:

% calculated based on total patients (n = 162).

Source: Medical records.

months). There was a highly significant statistical association between complications and an increase in the mean age of patients at diagnosis and initiation of treatment for congenital muscular torticollis ($p < 0.01$).

As for the median age, it was higher in patients with dentofacial complications of the disease (7.0 months) compared to the overall age (4.8 months) and in patients who did not present sequelae (4.0 months), with a statistically significant association of complications with the highest value of the median ($p < 0.03$). however, the association found between the median and the sequelae was lower than that of the mean age.

The mean age of onset of each of the dentofacial complications of the disease was verified in table 4, plagiocephaly was the earliest onset complication with a mean of 6.6 months (SD = 0.5), followed in order of appearance by facial asymmetry, ocular dystopia and auricular dystopia with means of 7.8 (SD = 1.2). 11.0 (SD = 1.5) and 11.1 months (SD = 1.8) respectively. The absence of malocclusion in this analysis was not due to null cells, but to the fact that it was only present in one patient in the study sample.

The order of temporal appearance of complications or cephalometric tracing is shown in Figure 1, which has its origin in the plain cranial x-ray anteroposterior view of one of the patients with dentofacial complications of the casuistry and in the results of table 4.

This shows the radiological signs and the average age of onset of 4 of the dentofacial complications. Considering the lower and upper limits, it can be stated with 95% confidence that in the present case the population mean between the earliest onset dentofacial complication (plagiocephaly) and the later onset (atrial dystopia) ranges from 6.1 to 11.9 months.

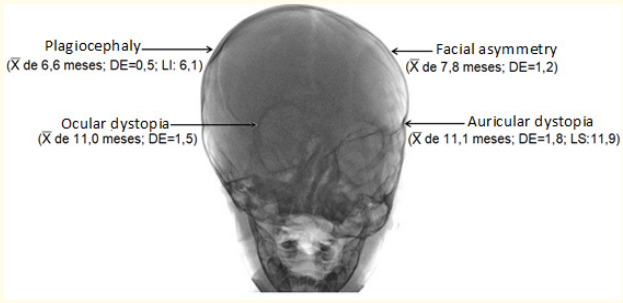

Figure 1: Cephalometric tracing of craniofacial complications

Discussion

Congenital muscular torticollis is known to be more common in boys than in girls [2,5,7,8]. however, in general there are not many studies where the sex variable is related to the complications of the disease; and the existing ones are usually contradictory, since the members of one gender or the other appear indistinctly as the most vulnerable [9,10].

The present research coincides with the results found by other authors, who in their studies found a predominance of males in patients with complications of this disease [11-13], although other authors suggest a predominance of girls [14,15].

The best-known complications of congenital muscular torticollis are dentofacial complications, although the spectrum of these is broader and also includes underdevelopment of the neck muscles, compensatory scoliosis and asymmetrical preference for the use of the limbs [4,5]. Gastroesophageal reflux disorder, psychomotor development, psychological and social dysfunction disorders such as poor academic performance, low self-esteem, depression, discrimination, nicknames and lack of opportunities for personal development have also been reported.[3,4] It should be noted that these complications were not the focus of this study.

The above explains the very diverse results obtained by different authors regarding the number and type of complications, although most of them agree in pointing to facial asymmetry and plagiocephaly as the most frequent [14-16].

The results of the present study are in agreement with the aforementioned studies regarding the higher frequency of plagiocephaly and asymmetry with respect to other dentofacial complications of congenital muscular torticollis, it should also be noted the low observance of malocclusion in the sample analyzed, with respect to this last finding the authors of the present research consider that it may be related to young age and the degree of bone maturity of the patients.

In most studies, age at diagnosis of congenital muscular torticollis and age at the start of rehabilitation treatment are unified as a single variable, as both are closely related [5,7,17-19].

In the present study, the association of complications with older age was found to be the diagnosis and treatment of the entity, which explains the general consensus on the importance of performing both early [20-24].

The authors of the present research believe that the late diagnosis of the disease and therefore the equally late initiation of conservative medical treatment in a significant number of patients, may be due to the fact that, unlike other diseases of equally congenital origin, there are no genetic or ultrasound markers for the prenatal diagnosis of congenital muscular torticollis, so its diagnosis is postnatal and fundamentally clinical. The latter can be difficult due to the characteristics of the neck of newborns and young infants (short and with little mobility).

It is necessary to emphasize this disease so that it can be detected by family doctors, pediatricians or parents as soon as possible, early and can be resolved in a simple and fast way through physical therapy and avoid delays in diagnosis that can obscure the therapeutic approach and compromise an evolution that a priori may be favorable [5].

Patients who are not diagnosed promptly develop craniofacial deformations and asymmetries with growth that include plagiocephalic deformation, zygoma retrusion, orbital dystopia, lateral deviation of the mandible, alteration of the orbital and occlusal plane and dental malocclusions, as well as a tendency to class II molar relationship on the side affected by torticollis and class III relationship on the opposite side [5].

Although the dentofacial complications of the disease are recognized, the age of their onset has not been fully clarified, as there are dissimilar criteria according to the different authors [5,9,25].

Regarding the above, Cueto., *et al.,* [5] place the appearance of facial asymmetry at an age as early as 6 months, while Tonkaboni., *et al.,* [25] place it at 5 years of age. Regarding plagiocephalic frontal deformity, mandibular abnormality with its functional effects and inclination of the occlusal plane, both authors agree that they appear from 5 years of age, later on maxillary abnormalities appear and finally orbital and auricular abnormalities [5].

The results of this casuistry coincide with the aforementioned authors regarding the order of appearance of dentofacial complications of congenital muscular torticollis, but they disagree regarding the ages of their appearance, the latter was relevant especially for plagiocephaly and ocular dystopia which occurred at ages much younger than that described in the aforementioned scientific literature; which, in the opinion of the researchers of this study, could be related to the accessibility of patients in Cuba to universal, free, specialized, continuous and early health services; guarantees of an earlier diagnosis of both the disease and its dentofacial complications.

As a limitation of the study, it was found that there was no national research that took into account the ages of the dentofacial complications of this disease, which makes it impossible to compare the results.

It is concluded that the increase in the average age at diagnosis and treatment of congenital muscular torticollis increases the risk of dentofacial complications.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

Author Contributions

- **Conceptualization:** Pablo Antonio Hernández Dinza.
- **Data curation:** Pablo Antonio Hernández Dinza.
- **Formal analysis:** Pablo Antonio Hernández Dinza. Yaimet Pérez Infante.
- **Research:** Pablo Antonio Hernández Dinza.
- **Methodology:** Pablo Antonio Hernández Dinza. Yaimet Pérez Infante.
- **Project management:** Pablo Antonio Hernández Dinza.
- **Appeals:** Pablo Antonio Hernández Dinza.
- **Supervision:** Pablo Antonio Hernández Dinza.
- **Validation:** Yaimet Pérez Infante. Pablo Antonio Hernández Dinza.
- **Visualization:** Pablo Antonio Hernández Dinza.
- **Writing - original draft:** Pablo Antonio Hernández Dinza.
- **Writing - revision and editing:** Pablo Antonio Hernández Dinza

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