DOI: https://doi.org/10.54393/pbmj.v6i12.971

PAKISTAN BIOMEDICAL JOURNAL

https://www.pakistanbmj.com/journal/index.php/pbmj/index

Volume 6, Issue 12 (December 2023)

Original Article

Frequency of Congenital Muscular Torticollis in Children

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ARTICLE INFO

Key Words:
Congenital Muscular Torticollis (CMT), Craniofacial Asymmetry, Muscle Function Scale (MFS), Rehabilitation.

How to Cite:
Surriya, S., Maqbool, N., Jamil, M., Amin, K., Sohail, W., & Ullah, F. (2023). Frequency of Congenital Muscular Torticollis in Children : Congenital Muscular Torticollis in Children. Pakistan BioMedical Journal, 6(12). https://doi.org/10.54393/pbmj.v6i12.971

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Received Date: 10th November, 2023
Acceptance Date: 30th November, 2023
Published Date: 31st December, 2023

ABSTRACT

Congenital muscular torticollis (CMT) is identified by unilateral contracture of sternocleidomastoid muscle (SCM), resulting in lateral flexion of neck with rotation of chin to the opposite direction. **Objective:** To determine frequency of CMT in children presenting at Children Hospital, Lahore. **Methods:** This descriptive study was conducted in the Department of Physical Medicine and Rehabilitation (PM&R), Children Hospital and University of Child Health Sciences (CH & UCHS), Lahore from June 2022 to December 2022 and included 179 children who reported at Department of PM&R with CMT. The data were collected using a self-constructed Torticollis questionnaire and analyzed using International Business Machines Corporation Statistical Package for Social Sciences Version 23 (IBM SPSS V-23.0). **Results:** Out of 179 patients, 103 (57.5%) were male and 76 (42.5%) were female. Associated problems like head tilt 102 (57.0%), rotation deficits 99 (55.3%), craniofacial asymmetry 67 (37.4%), fibrotic mass 68 (38.0%) and flat spot on either side of head 56 (31.3%) were seen. A significant association was found between age and torticollis present at birth (p-value=0.012). **Conclusions:** CMT is more common among children with age less than one year. Most of the cases were delivered by vaginal delivery with a history of infection and difficult delivery. It is important to investigate the long-term effects of continuing head tilt on cervical spine and secondary complications of CMT.

INTRODUCTION

CMT also referred as wry or twisted neck. Torticollis originates from the Latin words 'torquere' (twisted) and 'collum' (neck), it could be congenital or acquired. It is a postural musculoskeletal deformity that appears in neonates and infants during the neonatal period or shortly after [1]. It is distinguished by one-sided shortening and contracture or fibrosis of SCM resulting in head tilting towards the involved side, causing the neck to bend and chin turning towards the opposite side [2]. With a little male predominance, the incidence varies from 0.3% to 2.0% of births (Male: Female = 3:2) [3]. It affects the right side more frequently than the left side. This condition may coexist with other disorders such brachial plexus damage, plagiocephaly, craniofacial asymmetry, and hip dysplasia [4]. The etiology is unknown; however, it has been associated with intrauterine malpositioning or trauma during delivery, infectious disorders that affect the mother (such as sore throats, influenza and rheumatic fever), beriberi, radiation, hypothermia, and hereditary susceptibility [5, 6]. Another factor that increases a baby's risk of developing it is the use of forceps or suction during vaginal delivery. This places stress on the newborn's SCM and causes it to tighten, making it difficult for a baby to rotate his or her neck. CMT is most commonly classified

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into three categories: Postural Torticollis without any muscle stiffness or restriction to passive range of motion. Muscular Torticollis characterized by tightness of the SCM and restriction of passive range of motion. SCM mass or Pseudo tumor with swelling of the SCM and restricted passive range of motion [7]. Children are often sent to pediatric rehabilitation owing to CMT. The diagnosis principally depends on clinical and physical examination findings [8]. Goniometer is a reliable instrument in measuring range of motion. In CMT, ultrasonography can be used to examine muscle tissue, assess muscle fibrosis and check resolution with a color histogram that calculates the muscle's cross-sectional area and thickness [9]. Muscle Function Scale (MFS) is used to detect muscle function. Infants typically exhibit anomalies in functioning of the lateral flexor muscles of the neck [10]. The SCM is examined and strengthened using the lateral up righting response [11]. The treatment is nonsurgical in most cases, including parents or care-givers education, positioning, myofascial release, stretching and strengthening [12]. Conservative treatment has improved the condition of more than 90% of CMT patients [13]. Botox or surgical alternatives may be considered if conservative treatment failed [14]. But if left unattended, it may result in craniofacial asymmetry, cervical muscle scarring with limited head movement and compensatory scoliosis [5].

The lack of awareness, root cause, early diagnosis and treatment was among the main difficulties faced and are referred at later stages, compromising the prognosis.

As this topic didn't get any spotlight in past so the true prevalence and reasons are yet to be explored. Therefore, understanding and exploring such topic was the choice to be able to apprehend and cure it at earlier stages and raise awareness. This study aimed to determine frequency of CMT in children.

METHODS

This descriptive study was carried out among children diagnosed with CMT referred to the Department of PM&R, CH and UCHS, Lahore from June 2022 to December 2022. The participants of this study included 179 children diagnosed with CMT. Using the sample size formula:

$n = [Z\alpha/2/e]2(p)(1-p)$

$Z\alpha/2 = 1.96 \quad e = 2\% = 0.02$

$p = 1.9\% = 0.019$

$n = [1.96/0.02]2(0.019)(1-0.019)$

$n = 179$

The children with congenital muscular torticollis, both male and female gender are included, head tilt from 5 to 20 degrees, age of child below 4 years were included in the study. Children of age above 4 years, CMT associated with a neurological pathology, infants with congenital abnormalities of cervical spine were excluded. A self-constructed Torticollis questionnaire was used to collect data. The questionnaire was constructed using items from pre-existing literature. Eight out of ten questions were simple yes or no questions. The questionnaire included variables like age, gender, side involved and mode of delivery, history of infection or difficult delivery, head tilt, rotation deficits, craniofacial asymmetry or fibrotic mass present at birth and history of previous physiotherapy treatment. Parents of all patients were included in the research. After approval and written consent from professionals, this questionnaire was used. The data were entered and analyzed using IBM SPSS version 23.0. Categorical variables were expressed in form of frequency and percentages. Bar chart was used to display data. Chi-square Test was used to find the association between age and torticollis present at birth. A p-value of less than 0.05 was taken as statistically significant. Ethical clearance was obtained from ethical committee of The School of Allied Health Sciences, University of Child Health Sciences, Lahore (No. 1169/SAHS; Dated 11/08/2022).

RESULTS

The study was conducted at the Department of PM&R, CH and UCHS, Lahore. The data were collected using a self-constructed questionnaire. Basic demographics are demonstrated in Table 1.

Table 1: Baseline Demographics Features of Subjects

Variables	Category	N (%)
Gender	Male	103 (57.5)
	Female	76 (42.5)
Age(years)	0-1	92 (51.4)
	1-2	45 (25.1)
	2-3	29 (16.2)
	3-4	13 (7.3)

Out of 179 patients, 56 (31.3%) patients were delivered by vaginal delivery, 21 (11.7%) patients had been delivered by cesarean section, 54 (30.2%) patients by forceps and 48 (26.8%) cases were the breech presentation. About 54 (30.2%) patients were less than 1 year and 28(15.6%) cases were more than 9 months. The descriptive data show 119 (66.5%) patient's lesions were located on the right aspect of the neck, 56 (31.3%) of the patients developed lesion on the left side of their neck. Only 4 (2.2%) patients had bilateral lesions. Physiotherapy for IMT in the form of passive anatomical stretching, rotation and side flexion of the involved SCM was performed at the Red Cross Hospital. Appropriate manipulations were taught to the mother for use as a home program, and the child was followed up at regular visits. On such a treatment regimen, 60% of cases showed marked improvement requiring no further treatment, 36 % of cases dropout and 4% required surgical intervention. Of the 54 surgical patients operated on over a 23-year period, follow-up studies

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were conducted on 16 (30%) with a mean period of 10.5 years between operation and reassessment. Figure 1 shows all categorical variables.

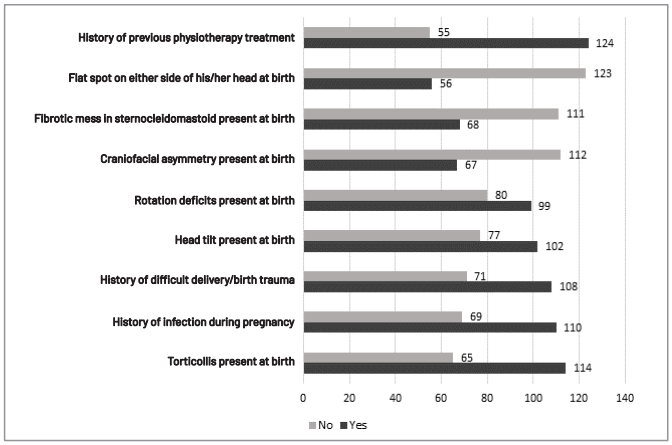

Figure 1: Bar Chart showing Variables under Study

DISCUSSION

The most frequent form of torticollis in children is CMT. It is worth noting that despite of numerous studies and no association of other clinical conditions to the course of torticollis, its etiology remains incompletely understood [15]. Numerous gynecologic and perinatal growth factors have been found to be associated with development of CMT [16]. In the literature, intrauterine mal positioning and trauma during delivery were responsible for CMT [17]. With this descriptive study, we have shown that CMT is an important reason for referral to Department of PM&R in the presence of breech presentation, difficult delivery and multiple gestation. We noticed slight male predominance with right sided preference of the CMT [18]. There were only four cases with bilateral lesions. Majority of the cases were delivered by vaginal delivery with a history of difficult delivery and infection during pregnancy. Associated problems like head tilt, rotation deficits, craniofacial asymmetry, and fibrotic mass and flat spot on either side of head at birth were seen. The retrospective study conducted by Amaral et al., in infants diagnosed with CMT referred to pediatric rehabilitation consultation [1]. The data suggested a male preponderance, which corresponds with our findings, however despite stated equal involvement of both sides, we found right side predominance in our study. Breech presentation during delivery was seen in majority of children with CMT. A palpable nodule was seen in 14 patients, whereas in our study 68 patients had fibrotic mass in SCM at birth. de Chalain et al., reviewed 54 cases of Idiopathic Muscular Torticollis (IMT) referred for surgery over a 23-year period and 134 cases referred for physiotherapy of which majority were less than 3 months of age. Demographic features, the role of physiotherapy, the timing of surgery and serial assessment were discussed and results were compared. Torticollis was present at birth in 50% patients which agrees with our study and more than 60% had involvement of the right side rather than the left. Male to female ratio was equal which contradicts our study [14]. The retrospective study by Sönmez et al., examined the data of patients treated for CMT in relation to age, gender, clinical characteristics, secondary deformities, site of the lesion, history of prior treatment approaches and screening procedures [5]. The findings revealed that the left side of the neck was involved more which contradicts our study and one case with bilateral lesion. Male to female ratio was equal. In our study male to female ratio was 103:76 in favor of boys and majority of the patients had lesions on the right aspect of the neck and four cases were seen with bilateral lesions. There was limited neck movement and related symptoms like tilting of head, craniofacial asymmetry and skull abnormalities in 50% patients as seen in our study. Eight patients presented with SCM tumor, whereas in our study 64 patients presented with SCM tumor. Das et al., conducted a retrospective case study among sixteen patients of congenital muscular torticollis [8]. Patient's age ranged from 5 days to 15 years, of which eleven were female and five were male, showing female predominance which contradicts our study as male predominance was found in our study. SCM was shortened in all cases (9 on the right side and 7 on the left side). Thirteen patients had the history of normal vaginal delivery, one patient had the history of forceps delivery and two had the history caesarian operation. In our study, 56 patients were delivered by vaginal delivery, 21 patients had been delivered by cesarean section, 54 patients by forceps and 48 cases were the breech presentation. Of 16 patients, 3 were neonates, 8 were infants, and 5 were more than 1 year of age. There was no associated anomaly. Out of 16 patients, 2 patients had a history of SCM tumor which agrees with our study. The treatment of CMT includes positioning, myofascial release, passive and active exercises, stretching of the tight SCM and strengthening of weak neck and trunk muscles [19]. Additionally, they can educate parents about torticollis symptoms and strategies to avoid complications [20]. Patients diagnosed at the age of less than one year can be treated effectively without any surgery. Surgical intervention should be regarded as a therapy of choice after the age of one year in order to avoid further long-term changes. The most typical surgical procedure is SCM muscle tenotomy.

CONCLUSIONS

Congenital muscular torticollis is the most common form of torticollis in children. Early diagnosis, parents/caregiver education and physiotherapy referral are prerequisite to achieve good results.

DOI: https://doi.org/10.54393/pbmj.v6i12.971

Authors Contribution

Conceptualization: SS
Methodology: MJ
Formal analysis: FU
Writing-review and editing: SS, NM, KA, WS, FU
All authors have read and agreed to the published version of the manuscript.

Conflicts of Interest

The authors declare no conflict of interest.

Source of Funding

The author received no financial support for the research, authorship and/or publication of this article.

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