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Demographic and Clinical Characteristics of Pediatric Patients with the Diagnosis of Torticollis: A Tertiary Clinic Approach

Tortikollis Tanısı ile Polikliniğimize Başvuran Pediatrik Yaş Grubu Hastaların Demografik ve Klinik Özellikleri: 3. Basamak Tecrübesi

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ABSTRACT Objective: The aim of this study was to evaluate the sociodemographic and clinical characteristics of the pediatric patients with torticollis admitted to our outpatient clinic. **Material and Methods:** Patients were evaluated retrospectively in terms of age, gender, congenital/acquired condition of torticollis, type of delivery, involved side, determined etiologic factors, applied treatments and concomitant clinical findings. **Results:** The mean age of our patients (36 females, 34 males) was 13.9±22.89 months (min: 1; max: 108). Right sternocleidomastoid muscle was involved in 57.1% of the patients. Sixty-four (91.4%) of the patients were diagnosed as congenital torticollis; while 8.6% (n: 6) was acquired torticollis. Of the cases with torticollis, 78.6% (n: 55) were born by normal spontaneous vaginal delivery and 12.9% (n: 9) were born by assistive methods such as vacuum /forceps. Physiotherapy exercises are the most commonly used treatment modality both for congenital and acquired torticollis. **Conclusion:** In pediatric patients with congenital or acquired torticollis, the basis of treatment is physiotherapy exercises. However, it is important to determine the etiology (birth trauma, intrauterine malposition, cervical hemivertebra, cervical lymphadenopathy, strabismus) and if possible, the underlying cause should be treated.

Keywords: Torticollis; congenital; acquired; exercise; ocular; surgery

ÖZET Amaç: Bu çalışmada kliniğimize başvuran pediatrik yaş grubu tortikollis tanılı hastalarımızın sosyodemografik ve klinik özelliklerinin değerlendirilmesi amaçlandı. **Gereç ve Yöntemler:** Tortikollis tanısı ile polikliniğimize başvuran hastalar retrospektif olarak değerlendirildi. Tedaviye başvuru yaşı, cinsiyet, hastalığın konjenital/kazanılmış olma durumu, doğum şekli, tutulan taraf, belirlenen etyolojik faktörler, uygulanan tedavi ve eşlik eden klinik bulgular incelendi. **Bulgular:** Hastalarımızın (36 kız, 34 erkek) yaş ortalaması 13,9±22,89 ay (min: 1; max: 108) idi. Hastaların %57,1'inde sağ sternokleidomastoid kası tutulmuştu. Olguların %91,4 (n: 64)'ü konjenital tortikollis tanısı alırken; %8,6 (n: 6)'sının yakınmaları kazanılmış olarak gelişmişti. Hastaların %78,6 (n: 55)'sı normal spontan vajinal yolla doğmuşken; %12,9 (n: 9)'u ise vakum/forceps gibi yardımcı yöntemlerle doğmuştu. Fizik tedavi egsersizleri hem konjenital hem kazanılmış tortikolliste en sık kullanılan tedavi yöntemi idi. **Sonuç:** Bu çalışmada, konjenital veya kazanılmış olarak gelişen tortikollisli pediatrik yaş grubu hastalarda tedavinin temelini fizik tedavi egzersizlerinin oluşturduğu ancak etyolojinin tespiti (doğum travması, intrauterin malpozisyon, servikal hemivertebra, servikal lenfadenopati, şaşılık gibi) ile beraber tedavinin nedene yönelik olarak yapılmasının gerekliliği vurgulanmaktadır.

Anahtar Kelimeler: Tortikollis; konjenital; kazanılmış; egzersiz; oküler; cerrahi

The term torticollis is derived from the Latin words tortus ("twisted") and collum ("collar" or "neck").[1]

Torticollis is the unilateral contraction of the sternocleidomastoid (SCM) muscles resulting in rotation of the head and neck with associated head tilt.

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The prevalence in the pediatric period is 0.4-1.3%; it may also develop as congenital or acquired.[1] Multiple causative factors have been documented in torticollis etiology, including muscular and nonmuscular causes.[2]

Congenital torticollis (CT) is the most common type of torticollis. Although the most common form of CT is congenital muscular torticollis (CMT); also neurological, ocular and cervical vertebral pathologies may cause the CT clinic.

The CMT is the third most common congenital musculoskeletal anomaly following developmental hip dysplasia (DHD) and congenital clubfoot.[3] The main pathological abnormality in CMT is fibrosis which develops by deposition of collagen and fibroblasts around muscle fibers leading to atrophy.[4]

Nonmuscular causes of torticollis are collectively not rare. In a child without an identifiable muscular etiology for torticollis; Klippel-Feil anomalies or an underlying neurologic disorder is likely to be the cause of the deformity in the majority of patients. About twenty-three percent of the causes of nonmuscular torticollis are of ocular origin.[5,6] Superior oblique palsy (SOP), the most common cause of ocular torticollis (OT) is 75% congenital.[5,7,8] Therefore congenital OT is frequently observed.

Acquired torticollis (AT) typically results from SCM or trapezius muscle injury or inflammation. It should be kept in mind that the AT may be a manifesting symptom rather than a disease in its own right. Fractures in the cervical spine, subluxations, osteomyelitis and tumors; infectious causes such as retropharyngeal abscesses, cervical lymphadenitis, tonsillitis and mastoiditis may lead to AT.[9]

In addition to early initiation of physiotherapy exercises in CMT; treatment success increases with correct positioning, environmental adaptive changes and close follow-up. Approximately 80-97% of patients with CMT do not require surgery.[4,10,11]

The treatment of AT depends upon the underlying cause. The correct assignment of the underlying cause is the basis of the treatment. History, physical examination, and cervical spine imaging will determine the etiology for AT in most of the patients. It is beneficial to add a suitable exercise program to the treatment of primary etiological factor in patients with AT.[12-14]

Limited number of studies about sociodemographic and clinical characteristics of the patients with torticollis have been observed in the literature. The aim of this study was to give an overview of the differential diagnosis of torticollis in children and to provide insight into our diagnostic and therapeutic algorithm.

MATERIAL AND METHODS

The approval of Tokat Gaziosmanpaşa University Faculty of Medicine Ethics Committee was obtained for our study (approval number: 03.04.2019/19-KAEK-098). Seventy patients (36 females, 34 males) who were admitted to our outpatient clinic between January 2013 and December 2018; under the age of 18 years; with the diagnosis of torticollis were included in the study. Patients with neurological and psychological diseases related with common muscle tonus anomalies were excluded from the study. The study was conducted in accordance with the principles of the Declaration of Helsinki.

The data of the patients were evaluated retrospectively from the electronic media files in the hospital automation system. In addition to the demographic characteristics of the patients (the age at the onset of the treatment, gender); type of torticollis (congenital, acquired); type of delivery (normal spontaneaous vaginal delivery, vacuum/forceps-assisted vaginal delivery, cesarean section); involved side (right, left); determined etiological factors (difficult delivery, breech presentation, cervical hemivertebra, cervical lymphadenitis, strabismus); applied treatments (only physiotherapy, torticollis surgery+physiotherapy, strabismus surgery+orthoptic exercises, torticollis surgery+brace+ physiotherapy); concomitant deformities (DHD, facial asymmetry) were recorded.

STATISTICAL ANALYSIS

Data analysis was performed by using SPSS for Windows 19.0 software program (IBM SPSS Statistics 19, SPSS inc., an IBM Co., Somers, NY). Descriptive statistics were given as number (n), percent (%),

median, mean and standard deviation. Comparing the median of the quantitative variables between groups, Mann Whitney U test and Kruskal Wallis test were used. *p* values below 0.05 were considered statistically significant.

RESULTS

The mean age of 70 patients included in the study was 13.9±22.9 months (min: 1, max: 108). Thirty-six (51.4%) of the patients were female and thirty-four (48.6%) of the patients were male. The mean age of the female patients at onset of the treatment was 12.58±20.32 months (min:1, max:72); while the mean age of the males was 15.29±25.58 months (min:1, max:108). There was no statistically significant difference between the onset of the treatment age by gender (p:0.335). The distribution of the patients by demographic and clinical characteristics is shown in Table 1. Sixty-four (91.4%) of our patients were diagnosed as CT; while six (8.6%) patients were AT. Three of the patients with congenital torticollis were due to strabismus and 2 had cervical hemivertebra. In the remaining 59 patients, the cause of torticollis was determined as CMT. In 57.1% (n: 40) of the patients' right SCM was involved; while 42.9% (n: 30) of the patients' left SCM was involved. There were no patients with bilateral involvement. In 12.9% (n: 9) of the patients, there was a history of a difficult delivery assisted by vacuum/forceps. Cervical hemivertebra was the most rare cause with 2 (% 2.9) patients. All 6 patients with AT were diagnosed with cervical lymphadenitis. These 6 patients were given exercise therapy in addition to antibiotherapy treatment for cervical lymphadenitis.

Exercise therapy was given to all patients with torticollis; on the other hand the number of patients treated with only exercise was 61 (87.1%). There was no concomitant deformity in patients with AT; 6 of the patients with CT had fascial asymmetry; 4 patients had DHD. Patients with facial asymmetry underwent surgery; 2 of these patients used cervical brace in addition to post-surgical exercise program. The distribution of the age onset of the treatment by applied treatment methods and concomitant deformities are shown in Table 2.

TABLE 1: The distribution of the patients by demographic and clinical characteristics

Variables	n (%)
Gender	
Female	36(51.4)
Male	34(48.6)
Type of torticollis	
Congenital	64(91.4)
Acquired	6(8.6)
Type of delivery	
Normal spontaneous vaginal delivery	55(78.6)
Vacuum/forceps-assisted vaginal delivery	9(12.9)
Cesarean section	6(8.6)
Involved side	
Right	40(57.1)
Left	30(42.9)
Etiologic factor	
Difficult delivery	9(12.9)
Breech presentation	7(10)
Cervical hemivertebra	2(2.9)
Cervical lymphadenitis	7(10)
Strabismus	3(4.3)
Absent	42(60)
Applied treatment	
Only physiotherapy	61(87.1)
Torticollis surgery+physiotherapy	4(5.7)
Diplopia surgery+ocular exercise	3(4.3)
Torticollis surgery+brace+physiotherapy	2(2.9)
Concomitant deformities	
Facial asymmetry	6(8.6)
Developmental hip dysplasia	4(5.7)
Absent	60(85.7)

DISCUSSION

Seventy patients with torticollis were evaluated over a 5-year period to ascertain the demographic and clinical characteristics of the disease in our study. And it was found that torticollis was seen in the same frequency in both genders, but CT was more frequent than AT. Some of our patients were born by cesarean section and all of the patients who developed fascial asymmetry underwent surgery.

Neurological, ocular, osseous, gastrointestinal and inflammatory causes should be considered in the differential diagnosis of torticollis.[5,11,15-17] In a retro-

TABLE 2: The distribution of the age onset of the treatment by applied treatment methods and concomitant deformities.

		Age onset of the treatment		
Variables	**n**	**Mean±SD**	**Median [Min-MaX]**	**p**
Applied treatment				
Only physiotherapy	61	9.72±17.23	3[1-60] (a)	0.011
Torticollis surgery+physiotherapy	4	42.4±43.65	36[1-108] (b)	
Diplopia surgery+ocular exercise	3	18±8.49	18[12-24] (b)	
Torticollis surgery+brace+physiotherapy	2	66±8.49	66[60-72] (b)	
Concomitant deformities				
Facial asymmetry	6	60.17±34.43	60[1-108] (a)	0.020
Developmental hip dysplasia	4	3±1.83	3[1-5] (b)	
Absent	60	10±16.47	3[1-60] (b)	

(ab): In same row, common letters indicate statistical insignificance.

spective study of 288 patients with torticollis, Ballock et al. found that 18.4% of the patients had non-muscular etiology.[6] In that group, they found different causes such as Klippel-Feil syndrome, brachial plexus injury, space-occupying lesions in central nervous system. In addition, torticollis due to ocular problems was also common.[6]

The majority of torticollis cases are congenital; a large part of them is known to be CMT with SCM fibrosis.[18] The etiology of CMT is still controversial. One of the hypotheses of CMT development is that the malposition of the head before delivery, causes injury and fibrosis in SCM.[19,20] Also there can be a familial predisposition.[21,22]

In our study, 91.4% (n:64) of the patients who presented with torticollis were CT, and %84.3 (n:59) of these were CMT. Our results are consistent with the literature. When our patients were evaluated in terms of etiology; difficult delivery assisted by vacuum/forceps (n:9); breech presentation (n:7), cervical hemivertebra (n:2), cervical lymphadenitis (n:6) and strabismus (n:3) were determined. In %60 of the patients, any underlying causes were not found. However, most of the patients were born by normal spontaneous vaginal delivery, it can be thought that birth trauma is the probable cause in patients without a determined etiologic factor.

Two of the 3 patients who had ocular torticollis were treated with strabismus surgery and 1 patient was treated with an orthoptic exercises regimen for strabismus. The application of early ocular surgery in children with OT, improves torticollis and prevents possible face and neck deformities.[8]

Determination of underlying causes is valuable in terms of providing treatment for etiology.[12,13] Surgery is still an important treatment option in patients with CMT accompanied by late onset of the treatment and facial asymmetries.[3] Different musculoskeletal pathologies such as craniofacial asymmetry, DHD, metatarsus adductus, clubfoot, C1-C2 subluxation and brachial plexus palsy can be observed simultaneously in children with CMT.[23-28] However, it is not clear whether these pathologies develop secondarily to CMT or due to common risk factors such as intrauterine malposition.

In our study, DHD was determined in 4 of the patients who were followed up with the diagnosis of CMT. In addition, craniofacial asymmetries were detected in 6 patients and surgical treatment was applied to these patients.

Without congenital-acquired discrimination; in the treatment of torticollis, physiotherapy applications are recommended not only for the patients receiving exercise therapy alone; for all patients who underwent surgery, who were using brace or who had both surgery and brace. Even, earlier initiation of physiotherapy is associated with increased effectiveness and shorter duration of therapy.[29-31] The goals of treatment for torticollis include achievement of midline head position, symmetric posture and gross motor skills; prevention/improvement of craniofacial asymmetry and resolution of restricted cervical range

of motion (ie, <5° limitation in active and passive cervical rotation and lateral flexion).[32]

In our study; while 87.1% (n:61) of the patients were able to reach the treatment goals with physiotherapy applications; 11,4% (n:8) of the patients underwent surgery. Two of these 8 patients, underwent diplopia surgery and 6 patients underwent torticollis surgery. The mean age of the patients who underwent surgery for CMT was 72 months (min:60; max:108). None of the patients with concomitant CMT and DHD underwent surgery. According to these results, concomitant DHD did not worsen the prognosis; the age at onset of the treatment could be more determinative in the need for surgery.

The main superiority of our study is its presenting a holistic assessment approach to pediatric patients with torticollis regardless of congenital-acquired discrimination. Thus, it was possible to determine the different causes of torticollis and the different treatment options.

LIMITATIONS

The present study is a retrospective cross-sectional study. The lack of data on antenatal history of our patients, such as birth week and birth weight and limited number of the patients due to being a single-center study are the main limitations of the study.

CONCLUSION

In a patient presenting with torticollis, it is important to determine whether the disease developed congenital or acquired. Especially in AT, if there is a possible etiological factor; it should be treated. With early initiation of physiotherapy programs; many patients; can be successfully treated with conservative methods without developing any deformity and need for surgery.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

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