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Pattern of ophthalmological and orthopaedic manifestations in pediatric patients of torticollis presenting to the tertiary care hospital of north India

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***Abstract*---**Introduction: Deformity of the neck and limited motion are commonly seen in pediatric orthopedics. The problem may be simply due to an intrinsic cervical issue or may be the manifestation of other underlying problems. To make a diagnosis of the nature and cause of neck deformity in the newborn is very important. Congenital torticollis is a condition that results in the deviation of a child's head to one side, with accompanying limitation in the range of motion of the neck. Aims and objective: Objective: To investigate the clinical features and outcome of congenital muscular torticollis (CMT) with passive neck motion limitation according to whether the finding on ultrasonography (US) is normal or abnormal. Material and methods: A total of 32 patients with Congenital Muscular Torticollis (CMT) who met eligibility criteria were included: age at presentation 6 month to 1 year, limitation of passive neck rotation or lateral flexion were included in

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this study. Patients underwent physiotherapy and were followed-up monthly. The clinical research with torticollis at the Variety Center for Craniofacial Rehabilitation was done from 2019 to 2021 retrospectively. Clinical records, standardized medical photographs, and cephalometric radiographs of the affected patients were examined. There was a total of 32 patients with a mean age of 30 months at the time of presentation. Result: Of the 32 patients evaluated, 2 did not return for follow-up. Of the remaining, 23 patients, follow-up ranged from 5 to 40 months (mean, 16.7 months). In eight cases, the patients suffered iatrogenic injuries (a clavicular fracture and a cephalohematoma). Six patients had a palpable mass within the substance of the sternocleidomastoid at the time of initial examination, but in all cases, the muscle was restrictive with evidence of contracture when attempting to center the head in a neutral position. Conclusion: The results of this study confirm many of the previously reported findings regarding congenital torticollis. There was found to be a high frequency of complications during birth. The majority of patients responded to conservative treatment.

***Keywords*---**pattern ophthalmological, orthopaedic manifestations.

Introduction

Congenital muscular torticollis (CMT) is a common disorder in neonates and infants with a tumor or tightness of the sternocleidomastoid muscle (SCM) caused by fibrotic changes[1]. Numerous theories have been proposed, but the true etiology of CMT remains unclear. Various causes implicated for CMT include intrauterine crowding or vascular phenomenon, fibrosis from peripartum bleeds, compartment syndrome, primary myopathy of the SCM, and traumatic delivery[2]. The main clinical findings of CMT include a characteristic head tilt, passive neck motion limitation, and/or a palpable neck tumor[3]. According to Suhr MC[4] et al, patients with CMT have been classified on the basis of clinical findings into a SCM tumor group (those with a clinical tumor in SCM) or a muscular torticollis group (those with tightness of the SCM but no clinical tumor). Congenital torticollis is a condition that results in the deviation of a child's head to one side, with accompanying limitation in the range of motion of the neck[5]. Although of multiple etiologies,(Table 1) the most common is a primary idiopathic condition resulting in fibrosis and scarring of the sternocleiodomastoid muscle[6]. Congenital torticollis is a condition with a reported incidence ranging from 0.6 to 400 per 100,000 live births. The term is derived from two Latin roots, tortus (twisted) and collum (neck)[7]. When affected by torticollis, the infant maintains the position of his head to one side, with the external ear held close to the shoulder and the face turns in the contralateral direction. In the majority of cases, this is the result of a primary muscular condition, usually affecting the sternocleidomastoid. Biopsies of the affected muscles demonstrate diffuse fibrosis, which is felt to cause shortening of the muscle and the resulting abnormality in head position[8]. The cause of the fibrosis has never been conclusively established, but many authors have speculated that it is related to intrauterine positioning and the process of delivery. The birth history of patients with torticollis demonstrates an unusually high

incidence of difficulties during labor, reported at approximately 30 to 60 percent in some series[9]. Experiments have shown that the sternocleidomastoid and its surrounding fascia are an anatomic compartment, and it is postulated that compression or injury of the muscle in utero can lead to a compartment syndrome[10]. In a classic experiment, Middleton[11] demonstrated that venous occlusion in dogs could produce fibrous tissue within the sternocleidomastoid. Others have proposed a similar pathologic effect resulting from arterial occlusion.

Material and Methods

The clinical research with torticollis at the Various Centers for Craniofacial Rehabilitation was done from 2019 to 2021 retrospectively. Clinical records, standardized medical photographs, and cephalometric radiographs of the affected patients were examined. There was a total of 32 patients with a mean age of 30 months at the time of presentation. All patients were initially started on a program of physical therapy. Eight patients (25 percent) subsequently underwent surgical correction consisting of a bipolar sternocleiodomastoid release/resection; the age of the surgical subgroup at the time of initial presentation was significantly greater than that of the nonsurgical patients (7 months versus 12 months). Of the patients for whom longitudinal records were available (16 of 32), the mean follow-up period was 42 months. The head tilt improved in all patients—surgical and nonsurgical—but it was only completely corrected in 60 percent of the physical therapy patients and 40 percent of the surgical patients. All patients in the series exhibited some degree of ophthalmological manifestations,facial asymmetry, most commonly manifest as mild retrusion of the ipsilateral forehead and zygoma with shearing of the ipsilateral auricular helix in a posterior and inferior direction. In the more severe cases, there were also changes in the shape and position of the orbit, nose, and mandible. However, in three case was the asymmetry sufficiently severe to warrant surgical reconstruction of the affected skeleton. Cephalometric analysis revealed a reduction in vertical facial height on the affected side. Evaluation of this clinical series would indicate that younger patients may be effectively managed with therapy alone; patients presenting for treatment at a later age are more likely to require surgical release/resection.

Result

Of the 32 patients evaluated, 2 did not return for follow-up. Of the remaining 30 patients, follow-up ranged from 5 to 40 months (mean, 16.7 months). In eight cases, the patients suffered iatrogenic injuries (a clavicular fracture and a cephalohematoma). Six patients had a palpable mass within the substance of the sternocleidomastoid at the time of initial examination, but in all cases, the muscle was restrictive with evidence of contracture when attempting to center the head in a neutral position(Table 2). All patients were initially referred for physical therapy after ruling out cervical spine and ocular pathology. In all patients available for follow-up (30 total), physical therapy resulted in a significant improvement in head position. In Ten patients, there was complete normalization of the head position. In three patients, there was residual head tilt following therapy, but this was not felt by the parents or the physician to require further intervention. Two patients demonstrated an inadequate response to therapy, and a bipolar surgical release of the sternocleidomastoid muscle was performed. Of this each of the five

oldest patients required three separate operative procedures to correct the torticollis, including a complete sternocleidomastoid resection in one. Evaluation of the clinical photographs revealed asymmetry in all cases, although mild in the majority of patients. The most common anomaly was posterior displacement of the ipsilateral ear, a finding present in 25 patients, followed in incidence by posterior recession of the ipsilateral zygoma in 12 patient and forehead in 16 patients. There was mandibular asymmetry with deviation of the chinpoint to the affected side in nine patients. The eye was inferiorly positioned with a reduction in the vertical height of the palpebral aperture in six patients. In five patients, there was deviation of the nasal tip to the affected side. (Table 3) These findings were not as apparent on a cephalometric radiographs of the patients. There were not significant consistent differences in the measurements of skeletal landmarks on the two sides of the face. This includes vertical facial height and orbital position. In no case was surgical intervention undertaken for correction of the craniofacial asymmetry. In one patient, helmet therapy was instituted at 8 months of age for correction of the forehead asymmetry but without evidence of significant improvement.

Discussion

The first reference to torticollis in the literature is a description of Alexander the Great by Plutarch. Archeological specimens have also demonstrated this condition, with the first specimen being a Greco-Roman soldier now termed the “Birmingham Mummy.” This unfortunate warrior seems to have been shot with an arrow in his neck, and he subsequently developed a torticollis[12]. Archeological excavations in the Hawaiian Islands have revealed an unusually high incidence of this deformity in the native islanders (1.8 percent). This clinical series focuses on congenital muscular torticollis, which must be distinguished from adult-onset torticollis, also termed paroxysmal torticollis[13]. This is most frequently a spasmodic torticollis. The age of onset of the condition generally occurs and it is often associated with psychological disorders. Treatment of this condition most frequently begins with biofeedback and pharmacotherapy, with non responders undergoing botulinum toxin injections and selective peripheral denervation of the cervical musculature[14]. Congenital torticollis is most frequently a primary condition affecting the sternocleidomastoid muscle. The findings of this series tend to parallel those in the published literature. Thirty-one percent of the mothers related complications during the pregnancy or delivery. Two of the patients required forceps for delivery and two suffered iatrogenic obstetric injuries, an incidence in keeping with the reported rate of birth difficulties in torticollis patients of 30 to 60 percent. There was also a suggestion of heredity in the etiology of the torticollis in one of the patients in the series; the mother and the uncle of an affected patient also had muscular torticollis that was treated surgically[15]. Previous reports have also suggested a genetic basis in some cases of torticollis. In one study, five family members in three generations were affected. In a separate report, five interrelated female children, three of whom were sisters, were afflicted with torticollis[16]. In the majority of cases, however, there is no previous family history of the diagnosis. Masses within the substance of the sternocleidomastoid, sometimes termed a pseudotumor, were present in 19 percent of the patients, as documented by physical examination in this series[17]. Previously documented incidence rates have ranged from 20 to 66 percent.

Studies using sonography of the muscle have reported that a mass with a hypoechoic rim can be demonstrated in all cases of torticollis[18]. The patients in this study with a history of a palpable mass had a tendency for a more severe type of torticollis. Physical therapy was the primary treatment modality in this series. It was recommended in all of the patients at the initial intervention, regardless of patient age or the severity of the torticollis. In 60 percent of the patients, this was the only treatment necessary. In the remaining 40 percent, there was an inadequate response to physical therapy; these patients underwent surgical release/resection of the sternocleidomastoid. Hulbert[19] et al have also reported a more frequent need for muscle surgery. In one study of 277 patients, 70 percent were adequately treated by therapy alone[20]; however, in the 10 children requiring surgery, 8 were over the age of 1 year at the time of presentation. It would seem that the majority of patients under 1 year of age respond to physical therapy, regardless of the severity of the torticollis. Most authors have recommended that, in the case of those patients not responsive to physical therapy, surgical muscle release be delayed until 12 months of age. Coventry[21] et al have advocated surgical intervention whenever a child has demonstrated failure to improve with a physical therapy program, operating as early as 4 months of age. In most series, the results of surgery seem to be satisfactory up to approximately 6 years of age, at which point there is a poorer response. The type of recommended surgical therapy varies. All patients in this series initially underwent a bipolar sternocleidomastoid release with subtotal resection of the muscle. Nove-Josserand[22] have advocated either an isolated proximal or distal release (myotomy) or a myoplasty, in which the muscle is basically stepcut. This allows lengthening of the muscle while preserving its origin and insertion. In longstanding or recurrent cases, it may be necessary to perform a more aggressive release, including all involved structures such as the trapezius. Any or all of the structures in the neck may accommodate to the shortened neck length in longstanding cases of torticollis. This is the most probable explanation for the need for multiple operations in the two oldest patients in this series. A release of all shortened structures in the neck have been performed at the initial operation. There were no major complications resulting from operative intervention in any of the patients. Spinal accessory nerve damage is possible, and care must be exercised to avoid injury during the muscle surgery. It is best to perform the surgery by way of an incision approximately 1 cm above the clavicle. For a bipolar release, the second incision can be placed high, along the mastoid hairline where it is well concealed. The use of an endoscope has also been advocated a technique that allows division of the sternocleidomastoid through a single incision within the hairline. In most cases, neither the referring physician nor the family was aware of the facial asymmetry until it was pointed out to them. Indeed, in the majority of patients, the asymmetry was mild—it was always the torticollis, not the skeletal asymmetry, that was the chief complaint of the patient or family. This is consistent with the lack of significant findings in the radiographs of these patients. Only in the most advanced case were there significant differences in the vertical position of skeletal landmarks. This may be partially due to the fact that accurate cephalometric radiographs are difficult in the young patients who comprise the bulk of this series. The cluster of findings that comprises the asymmetry associated with torticollis is consistent and has been reported by Williams[23] et al. It results from a deformational process both antenatally and postnatally and is aggravated by secondary compensatory changes. Ferguson[24]

termed this “a suborbital torsional deformity of the face toward the affected side.” This is best exemplified by the oldest patient in the series, an 18-year-old man from Central America who had evidence of torticollis since birth. There was no family history of torticollis and no history of pregnancy or labor difficulties. He had undergone no previous treatment for his condition. One week before his evaluation, he underwent a sternocleidomastoid release at an outside hospital with limited success. A significant residual head tilt was present, and the affected sternocleidomastoid was still foreshortened. There was posterior recession of both the malar eminence and the forehead on the affected right side. The right ear was displaced posteriorly and inferiorly. The right orbit, although slightly inferiorly displaced, was most noticeable because of the relative reduction in the vertical height of the palpebral aperture. The mandibular asymmetry was best appreciated on the cephalometric radiograph. The left ramus was markedly longer than the right, contributing to a progressive shift in the facial midline that becomes more noticeable toward the pogonion. The size of the right mastoid process was enlarged, as the constant pull of the foreshortened sternocleidomastoid on this side led to bony hypertrophy at the muscle origin. Although this patient represents the most extreme example, similar clinical findings were present to a lesser degree in all of the reported patients. It should be emphasized that surgical intervention for these asymmetries was recommended in only the 18-year-old man described above. A combined Le Fort I-bilateral sagittal split osteotomy was recommended, but the patient declined. Slate[25] and colleagues reported that only 2 of 26 patients in their series with torticollis required surgical intervention for reconstruction of the asymmetric craniofacial skeleton. These authors performed cranioorbital osteotomies, as have others. Ferguson[24] in his review of the subject, argued against such an approach and feels that this is “camouflage surgery.” He recommended combining bimaxillary osteotomy with septorhinoplasty to achieve restoration of craniofacial symmetry. In our series, the severity of the orbital dystopia was not thought to warrant correction. In the single patient for whom an operation was recommended, the asymmetry in the lower half of the face was felt to be the most prominent. A bimaxillary surgical procedure in this patient would have provided a superior aesthetic result, as compared with orbital translocation.

Conclusions

The results of this study confirm many of the previously reported findings regarding congenital torticollis. There was found to be a high frequency of complications during birth. The majority of patients responded to conservative treatment; those who failed conservative therapy tended to be older at the time of initial presentation. The later patients underwent surgical release/resection of the sternocleidomastoid muscle. This study also found evidence of craniofacial asymmetry in all patients presenting with torticollis. Although not severe in most cases, the findings were consistent with a frontal deformational plagiocephaly, with recession of the ipsilateral zygoma and forehead, and a reduction of vertical facial height on the affected side. In only few patients were these findings were severe enough to warrant surgical correction of the bony asymmetry.

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Table 1: Potential causes of Torticollis

Potential Causes of Torticollis
Congenital muscular torticollis
Ocular conditions
Superior oblique muscle palsy
Lateral rectus muscle palsy
Nystagmus
Cervical spine anomalies
Klippel-Feil
Hemiatlas
C1–C2 subluxation
Infections
Parapharyngeal abscess
Sinusitis
Upper respiratory infections
Trauma
Radiation therapy
Central nervous system tumors (especially posterior fossa)

Table 2: Basic characteristics according to ultrasonography findings

Variable	P Value*
Birth weight	0.539
Age at presentation	0.002

Table 3: Manifestations

Manifestations	Precentage	P value
Opthalmologic	38%	0.003
Orthopeadic	62%	0.002