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One Step Tenotomy in Congenital Torticollis: A Case Report

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Abstract

BACKGROUND: Congenital muscular torticollis (CMT) is a common pediatric abnormality involving sternocleidomastoid (SCM) muscle. The pathogenesis of CMT has yet to be elucidated, but intrauterine abnormalities seem to be a plausible explanation. This condition, if left untreated, may result in craniofacial asymmetry, neck pain, and limited neck movement. We reported a case of an 11-year-old girl with CMT, which symptoms were first noticed at the age of 4, but were neglected by her parents, the patient was then treated with a complete unipolar SCM release.

CASE PRESENTATION: An 11-year-old girl presented with neck stiffness and limited head movement, first noticed at the age of 4, but was neglected. These symptoms worsened within the last few weeks. Physical examination revealed tension and tightness in the left SCM muscle. The patient then underwent complete unipolar release of SCM muscle and proceeded to aggressive physiotherapy for 3 months and put to rigid collar neck for the next 3 weeks. Follow-up was done in the fourth week and a full range of motion (ROM) of the neck was achieved.

DISCUSSION: The timing of surgery could yield a good result if performed within 1–4 years of age. However, another literature stated that surgical intervention in the older patient with CMT could still result in better outcomes. Therefore, in this case, we proceeded to perform a complete unipolar release on the left SCM muscle. Post-operative results were satisfying, with significant improvement in neck ROM.

CONCLUSION: Surgical approach in adolescents with CMT may still carry a favorable outcome, in terms of better ROM and neck motion. Subsequent physiotherapy and brace placement were crucial to maintaining the results of the surgery.

Introduction

Congenital muscular torticollis (CMT) is a condition that involves sternocleidomastoid (SCM) muscle to be contracted, which is usually presented at birth or shortly after birth [1]. The term “torticollis” originated from Latin words: Tortum collum, meaning twisted neck. This terminology was first described by Tubby in 1912, as “a deformity, either congenital or acquired, characterized by a lateral inclination of the head to the shoulder, with torsion of the neck and deviation of the face” [2], [3].

CMT is the third most common congenital musculoskeletal anomaly, after hip dysplasia and clubfoot [4], [5]. Historically, the reported prevalence of CMT ranged from 0.3% to 2% [6] and as high as 3.92% in neonates [6]. The incidence of infants affected with CMT is 3.9% to 16% [7].

CMT is a common condition caused by shortening, thickening and/or tightness of the SCM muscle, mostly unilateral, characterized by fibrosis in histologic finding [2], [6]. Ipsilateral cervical lateral flexion and contralateral cervical rotation, followed by SCM mass, are the clinical features of CMT [6]. In 50% of CMT cases, SCM mass do not appear. This condition is usually first identified in age 2 to 3 weeks as neonates and can persist until 1-year-old [3].

The cause of SCM fibrosis remains unclear. Some authors proposed that the occurrence of this condition is caused by at-birth/intrauterine compartment syndrome, intrauterine crowding and malposition, fibrosis due to peripartum bleeding, and primary myopathy [7], [8]. Other literature suggests the so-called “mesenchymal theory”, which is related to environmental change and results in the dedifferentiating of mesenchymal cells [9]. Other literature stated that around 30–60% of CMT cases demonstrated a history of difficulty during labor. A study by Kim *et al*. [4], found that approximately 16.6% of CMT patients presented in the breech position during labor, compared to the general population (3%–4%). It is hypothesized that the breech position of the fetus contributes to the shortening and fibrosis of SCM muscle [10], [4].

The primary pathologic finding of CMT in the involved SCM muscle includes excessive endomysial and perimysial fibrosis, adipocyte hyperplasia, and muscle fiber atrophy [6]. These condition results in tightness and limited cervical motion. The worsening of the fibrosis is proportionately related to the increasing age of the child. Ultrasound is the radiological choice in assessing CMT. Careful evaluation should be done

to differentiate congenitally and acquired types or torticollis, for example, post-traumatic conditions, infection, inflammation of adjacent structures, tumors, and ocular torticollis [11].

Most cases of CMT resolve completely either spontaneously within months after birth or with a conservative approach. Early initiation of conservative treatment (<1-year-old), such as physiotherapy and gentle controlled passive manual stretching exercises, yielded excellent results in up to 90% of cases [9], [12]. Surgical intervention is usually preferred in older patients, patients with residual head tilt, passive rotation deficit, or lateral bending of more than 15° at the age of 6 months. Surgical approaches include unipolar or bipolar release of SCM muscle, thigh fibrous band release, release with Z-plasty, endoscopic release, or muscle resection. The timing of surgery in CMT is still a matter of debate. Several literatures have suggested early release to avoid craniofacial asymmetry and to reduce the length and expense of treatment. Surgical treatment after the age of 1 year has been advised, to allow post-operative recovery time [2], [8].

The disadvantages of early surgical intervention were incidents of more frequent wound breakdown, formation of hematoma, and superficial wound infection. Here, we present a case of a girl with the left-sided CMT, diagnosed at age of 11 and underwent unipolar release of SCM muscle, followed by 3-weeks of rigid collar neck usage.

Case Presentation

An 11-year-old female child presented with the complaint of neck stiffness and limited head movement. These symptoms were first noted at the age of 4 but were neglected by the parents. Initially, the patient was not hampered by the symptoms, but the symptoms started to worsen during the past few weeks. The mother's gestational history was not significant. The history of trauma during labor was denied by the parents. The patient was born by cesarian section at 38 weeks of gestation due to breech presentation. She had 3100 g of birth weight and 48 cm of birth height. Growth and development were within normal limits. Immunization status was completed and no relatives or family with a similar condition.

On physical examination in resting position, the patient's head was tilted to the left and chin deviated to the right (Figure 1). There was a significant restriction of neck movement, especially on lateral rotation. Facial asymmetry was not documented. On palpation of the left SCM muscle, it was a non-tender, taut, and cord-like sensation. No lump or mass could be palpated along the entire SCM muscle. Radiographs of the cervical spine, hips, and lower extremities were normal. No neurological deficit was noted.

Figure 1: Pre-operation clinical photograph showing a female patient with congenital muscular torticollis affecting left sternocleidomastoid muscle, lateral, and anterior view

Based on the above clinical features and physical examination, a diagnosis of CMT was established. The patient was then scheduled and underwent left-side unipolar SCM release. The surgery was performed by an experienced pediatric orthopedic surgeon. In this procedure, we performed a complete release of both the sternal and clavicular head of the SCM muscle. We did complete release until perivascular fat surrounding the carotid sheath. Then, we did an excision of the distal part of the SCM muscle approximately 0.5 cm to ensure; there was no bridging fibrosis between the SCM muscle to the clavicle and sternal regions which would lead to suboptimal correction (Figures 2 and 3).

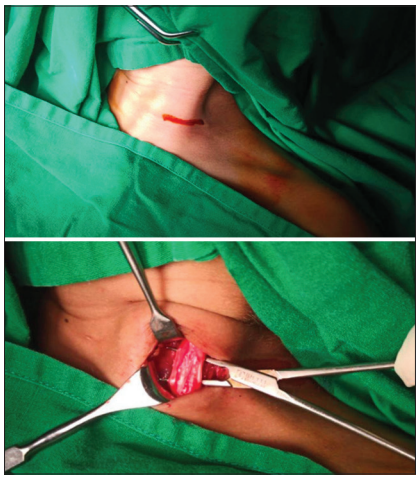

Figure 2: Intraoperative photographs show incision marking (upper) and surgical release/tenotomy of the left sternocleidomastoid muscle

Postoperatively, the patient was put on a rigid collar brace for 3 full weeks and aggressive physiotherapy was commenced for the next 3 months. Follow-up of the patient was expected 4 weeks after the surgery.

Four-week follow-up of the patients found that there is an improvement in the range of motion (ROM) of the neck, which is to the left and right (Figure 4). Physiotherapy was continued until the next 8 weeks, and followed by a home exercise regimen once the child was discharged from therapy.

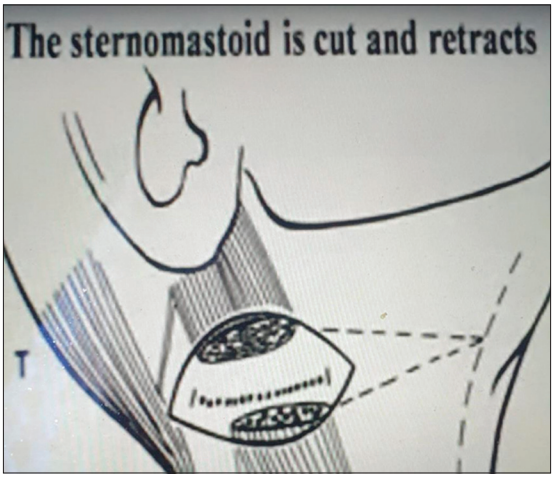

Figure 3: The picture above shows an illustration of the surgical release of the sternocleidomastoid muscle

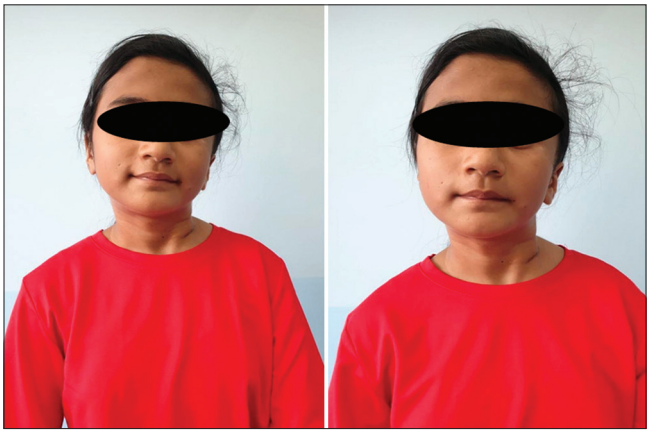

Figure 4: The photographs above were taken 4-week post-operation

Discussion

CMT or "wry neck," is described as painless torticollis and is among the most common musculoskeletal deformities in children. It is presented by a persistent head tilt toward the affected side, with the chin rotated in the opposite direction [4]. The twisted neck position of the neck may lead to positional plagiocephaly or asymmetry craniofacial. This condition may be associated with hip dysplasia or metatarsus adductus. Most of the literature describing CMT is found in orthopedics journals due to its effect on the cervical spine and its association with hip dysplasia. However, most patients are often referred to otolaryngologists for the evaluation of "neck tumor." To counter this, careful utilization of imaging techniques such as ultrasound should be considered to diagnose CMT [10]. As in our case, the patient did not complain about the neck mass, instead was the stiffness of the neck, which increase the likelihood of an orthopedic department visit.

The reported incidence of CMT ranges from 0.3% to 2% with male predominance (3:2) [5], [13]. The right SCM muscle is more often affected than left. Plagiocephaly is reported in up to 90% of children with CMT [7]. The etiology of CMT is still unclear [9], [14]. Birth trauma, prenatal or postnatal compartment syndrome, and impairment of the developing SCM due to intrauterine constraint were the most promising pathophysiology explanation [6].

Treatment of torticollis depends on the age of the patients, the severity of torticollis, the absence of plagiocephaly, and the presence of associated neuromuscular or orthopedic impairment [7]. At present, there are no standard guidelines for the treatment of torticollis. In general, treatments range from physical therapy (conservative), using orthoses and surgical release. Approximately 50%–70% of SCM mass resolves spontaneously during the first year of life with minimal residual deficits. Conservative intervention may be first recommended in younger CMT patients and yield up to a 90% success rate [5], [15]. The study by Keklicek and Uygur [16] showed that home programs which consisted of positioning the neck and head, handling strategies, stretching exercises and environmental adaptations were effective in managing CMT.

In children older than 1-year-old in which conservative management failed, a surgical approach should be considered as an option to prevent irreversible changes. Immediate surgical release of SCM muscle should be done to obtain satisfactory results [13]. There were various surgical procedures reported for the management of CMT which include unipolar SCM muscle lengthening, bipolar SCM muscle lengthening, Z lengthening, or radical resection of the SCM [7]. The procedure chosen is determined by the surgeon's preference and the amount of SCM tightness. The bipolar release was usually done in older patients and with more severe deformity [2], [13]. Many authors suggested using the bipolar release approach to treat CMT in the older population as it yielded good outcomes [5], [8], [17], [18], [19]. However, others proceeded to use the unipolar approach due to various reasons [2], [13]. In our case, we chose to undergo unipolar SCM release to correct the deformity as a concern of injury of both facial and spinal accessory nerves during the release of SCM muscle from its mastoid attachment.

Concern rises in our surgical approach in this patient, as the clavicular head could reattach to the clavicle and form a lateral band. This condition may require secondary release operation [13]. As reported by Chotigavanichaya *et al*. [9], there were 26.5% cases of recurrence after unipolar release.

The timing of surgery is controversial. Ling [17] stated that the optimal time for surgery is between 1 and 4-year-old. Other literature also supports that full recovery of facial asymmetry after the age of 4 is difficult to achieve [20]. However, several authors reported that

late release of SCM muscle for patients >6 years old could yield an acceptable result.

In adults with CMT, surgical options were often limited and the complication rates were high. Some clinicians used the bipolar release technique, which may improve the functional aspect of neglected CMT. Other outcomes using this approach may include complete muscle release, satisfactory cosmetic appearance, and minimal recurrence [5], [18]. Efficacy of bipolar release was also assessed by Seyhan *et al*, which found an improvement in ROM (10° to 25°) in most adult patients (mean age 14.6 years old). It has been thought that surgery may not be favorable in patients aged 10 or older. Nevertheless, surgery may still help to improve neck motions and correct head tilt [19].

In other articles, the unipolar release approach was used in adolescents (mean age of 15) as reported by Kamboh *et al*. The authors reported up to 80% yielded excellent or good outcomes, which were assessed with modified Lee's score. No significant complications and recurrences were observed in all patients [10]. The downside of unipolar release is recurrence could be as high as 7% [11], [19]. Hence, because in our case, we proceed to undergo unipolar release, emphasizing in post-operative physical therapy compliance is substantial.

Although early surgical release often yielded significant results in neck ROM improvement, delayed intervention is not uncommon among CMT patients. Inadequate treatment of CMT may result in complications such as persistent pain, spinal deformities, and craniofacial abnormalities [5]. Nevertheless, a study from Min *et al*. [5], showed that even in an older patient (> 5-year-old), surgical release is still effective in terms of spinal deformity improvement. Kamboh *et al.* also reported improvement in cervical-mandibular angle (19.62 ± 7.06 vs. 14.03 ± 6.87; $p<0.05$) in 28 adolescent CMT patients [18]. A case series of 12 adolescent-adult patients reported by Patwardhan *et al.,* found that surgical intervention could improve pre-operative rotational and lateral flexion deficits ($p < 0.001$) [9].

The post-operative immobilization protocol for CMT is controversial. In the initial days following surgery, the patient tends to keep the head in its former position to reduce pain and compliance with the prescribed exercises is poor. If the head remains in this position, the released structures will regain their former tightness. Immobilization in an overcorrected position has been claimed to give better results. Other literature suggested the long-term use of braces to prevent recurrence [1]. Cheng *et al*. stated that post-operative physical therapy is crucial in maintaining the surgery effects [14]. In this case, our patient complied with the physiotherapy exercise prescription and we prefer using a rigid collar neck brace to further immobilize the neck to prevent the recurrence of torticollis.

Conclusion

CMT is a rare condition, involving shortened or tighten SCM muscle, either unilateral or bilateral. This condition is caused by fibrosis of unknown cause, affecting primarily newborn infants or young children. Conservative management should be started after the initial recognition of the disease, preferably in children <1-year-old as it yielded high satisfactory results. Surgical intervention should be commenced in more severe cases or if failure in conservative management, to prevent further deformity (e.g., plagiocephaly). In neglected CMT cases, the bipolar release technique could yield favorable results, as well as unipolar release surgery. The important note is to perform a thorough surgical release until the carotid sheath is within the vicinity. Our proceeded procedure is generally well-tolerated with no major complications. Post-operative management such as physical therapy and immobilization is crucial to enhance the benefit of surgery.

Author Contributors

John Butarbutar, MD PhD – Conceptualization, Methodology, Validation, Investigation, Formal Analysis, Data Curation, and Writing – Review and Editing. Robby Oscar Sitohang, MD – Validation, Formal analysis, Data Curation, Writing – Original Draft, and Writing Review and Editing

Ethics Approval and Consent to Participate

The participant has been given written informed consent. There was no experiment on humans, so ethics did not involve in this study.

Consent for Publication

All authors approved the final version of the manuscript

Availability of Data and Materials

Data are available at the reasonable request

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