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Case report

A rare case of 9 years congenital muscular torticollis treated with complete unipolar sternocleidomastoid release: A case report and literature review

Aryadi Kurniawan *, Anissa Feby Canintika

Department of Orthopaedics and Traumatology, Faculty of Medicine, Universitas Indonesia, Cipto Mangunkusumo General Hospital, Jakarta, Indonesia

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ABSTRACT

Introduction: Congenital muscular torticollis (CMT), when left untreated, may result in numerous complications such as eye movement disorder, craniofacial asymmetry, neck pain and compensatory asymmetrical spine curvature that worsens with age. We reported a 9-year-old boy with neglected CMT treated with complete unipolar sternocleidomastoid release.

Presentation of case: A 9-year-old boy presented with his head tilted to the left since he was born. Physical examination revealed tense and uptight sternocleidomastoid muscle. The patient then underwent complete unipolar stenocleidomastoid release. Postoperatively, the patient completed 3 months of aggressive physical therapy thrice a week followed by a home exercise regimen once the patient was discharged from therapy. At six months of follow-up, the active range of motion of his neck was markedly improved in all directions, and his cervical spine was noted to be straight. The CMA was also decreased from 14 to 0 degree.

Discussion: It is recommended that, in those with CMT, the surgery should be performed between 1 and 4 years of age. However, in our case, the patient was already 9 years old. Nevertheless, he had achieved regained full cervical range of motion, and significant improvement of CMA angle (from 14 to 0 degree postoperatively) after undergoing unipolar sternocleidomastoid release and three months of aggressive physical therapy.

Conclusion: Surgical intervention followed by aggressive physical therapy for patients with neglected CMT, despite late presentation, may still carry a favourable outcome.

1. Introduction

Congenital muscular torticollis (CMT) is defined as a thickening and/or tightness of the unilateral sternocleidomastoid muscle (SCM) characterized by fibrosis, resulting in a shortening of the SCM and consequent limited neck motion [1]. When left untreated, CMT may result in progressive limitation of head movement which may end up in eye movement disorder, craniofacial asymmetry, neck pain and compensatory asymmetrical spine curvature that worsens with age. Recommended treatments starts with conservative stretching and splinting which can be routinely performed as an active home program under supervision. Surgical management of CMT has to be considered when the deformity is not corrected after the age of 1 years. Most surgeons agree that the prognosis is better if surgery is performed between the age of one to four years of age [2]. The prognosis is known to be influenced by the age at the time of surgery; thus, early diagnosis and treatment are important [3].

Neglected CMT is defined when the patient's condition remains the same after sessions of stretching and splinting and intervention initiated after age of four-year-old [2]. We reported a 9-year-old boy with neglected CMT treated with complete unipolar sternocleidomastoid release. The work has been reported in line with the SCARE criteria [4].

2. Case report

A 9-year-old boy presented with his head tilted to the left since he was born along with a lump on his right neck. The mother then brought her child to a pediatrician; she was told that the child had contracted muscle. The patient then had physiotherapy, and the lump started to disappear and the tilted head appeared better. However, as the patient got older, the tilted head became more severe. There was no pain. History of trauma, seizure, fever, and infection was denied as well. History of other congenital anomalies was denied. At the age of 9 years, there was no visible neck mass and neurological deficit, however there was a tense and uptight sternocleidomastoideus muscle (Fig. 1). Preoperative radiograph demonstrated cervicomandibular angle of 14 degree (Fig. 2).

* Corresponding author.
E-mail address: aryadi.kurniawan@ui.ac.id (A. Kurniawan).

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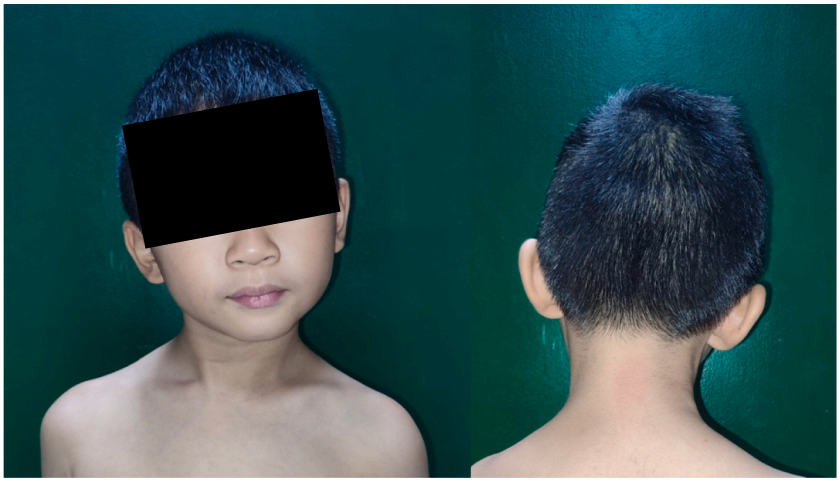

Fig. 1. Preoperative clinical picture demonstrating a shortened right sternocleidomastoid muscle (SCM) with associated tilting of the head to the right as well as rotation of the face and chin to the left.

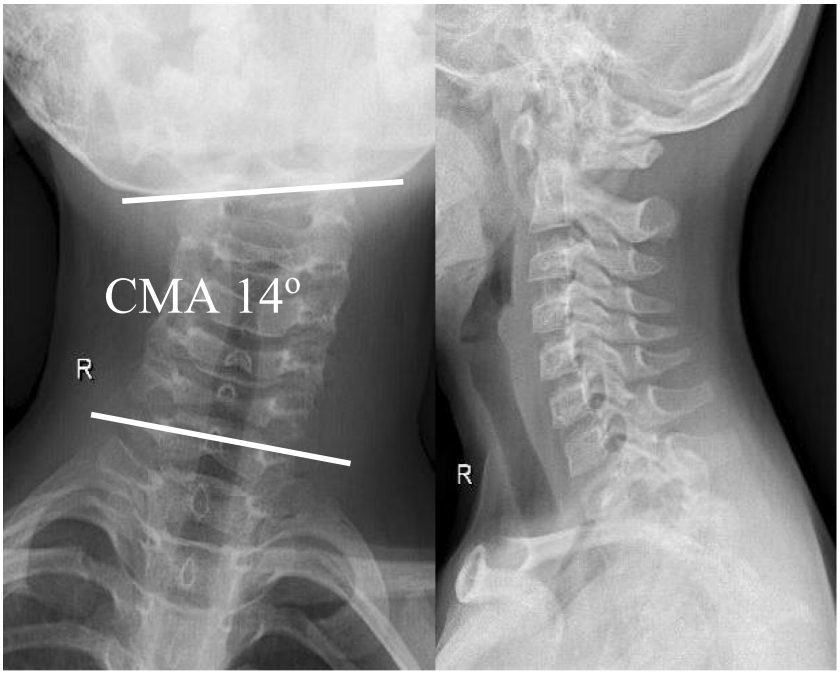

Fig. 2. Preoperative radiograph demonstrated no discontinuity and cervicomandibular angle of 14 degree.

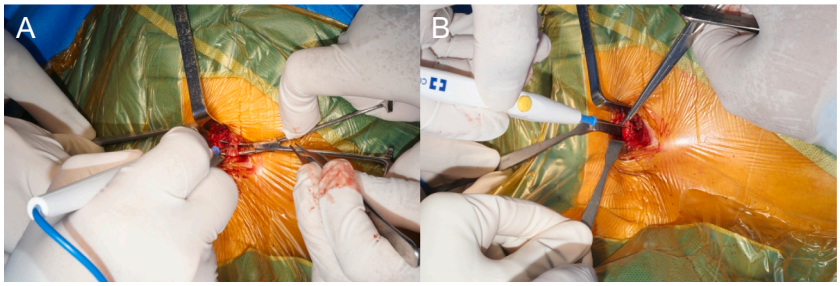

Fig. 3. (a) Complete release of sternal head of stenocleidomastoid muscle. (b) Complete release of clavicular head of stenocleidomastoid muscle.

During gestation, there was no alcohol consumption nor drugs overuse taken by his mother. History of trauma during delivery was denied. The patient was born by caesarean section at 38 weeks gestational age due to breech presentation. He had 3200 g of birth weight and 48 cm of birth height. The patient could sit at 8 months old, stand at 14 months and walk at 20 months. The patient had done

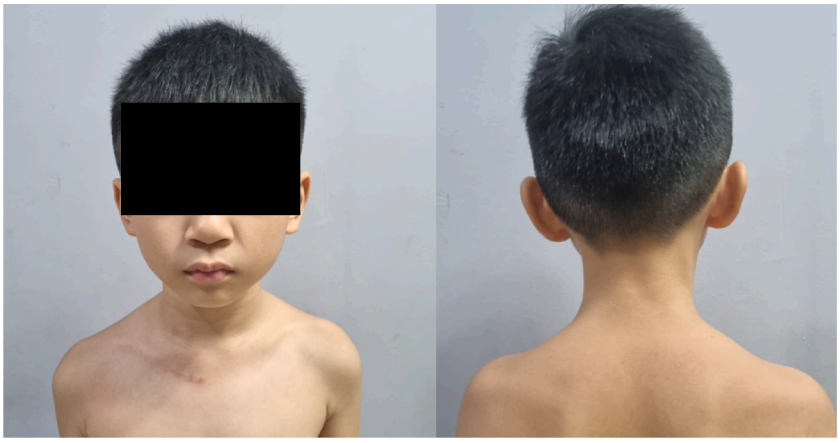

Fig. 4. Six months postoperatively, the cervical spine was in midline position.

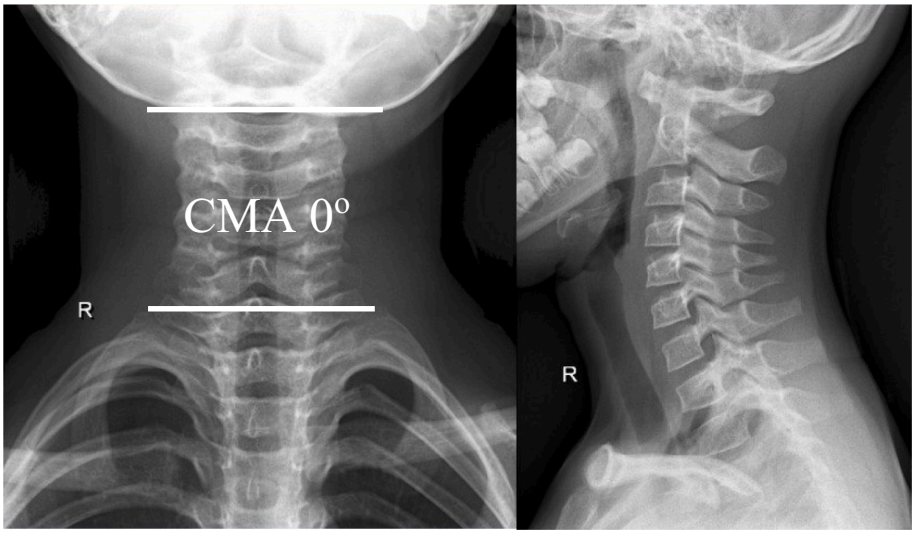

Fig. 5. Six months postoperative radiograph showed cervicomandibular angle of 0 degree.

complete immunization. There was no history of similar disease in her family.

The patient then underwent unipolar sternocleidomastoid release. This surgery was performed by an experienced pediatric orthopedic surgeon. In this procedure, we performed complete release of sternocleidomastoid muscle of both sternal and yang clavicular heads (Fig. 3). We did complete release until perivascular fat surrounding the carotid sheath. Then we performed excision of the distal part of sternocleidomastoideus approximately 0.5 cm proximally to ensure that there was no bridging fibrosis between the sternocleidomastoid muscle to clavicular and sternal regions which would lead to suboptimal deformity correction.

Postoperatively, the patient completed 3 months of aggressive physical therapy thrice a week followed by a home exercise regimen once the child was discharged from therapy. At six months of follow-up, the active range of motion of his neck was markedly improved in all directions (Fig. 4), and his cervical spine was noted to be midline. Both patient and parents were highly satisfied with the result. The CMA was also decreased from 14 to 0 degree (Fig. 5).

3. Discussion

Torticollis in Latin means twisted neck. Such terminology was first defined by Tubby in 1912 as "A deformity, either congenital or acquired, characterized by lateral inclination of the head to the shoulder, with torsion of the neck and deviation of the face" [5]. CMT is used to describe a condition presenting at birth or shortly after birth in which the sternocleidomastoid muscle (SCM) is shortened on the involved side, leading to an ipsilateral tilt of the head and a contralateral rotation of the face and chin. If inadequately treated, cervical muscles fibrose resulting in progressive limitation of head movement and craniofacial asymmetry, compensatory scoliosis, plagiocephaly, and elevation of the ipsilateral shoulder that worsens with age [6].

The reported incidence of CMT ranges from 0.3 % to 2.0 %. There is a moderate male predominance of approximately 3:2, and the right SCM is more often affected than the left [7]. The etiology of CMT is still a topic of debate. The oldest and most commonly accepted theory was initially proposed by Whitman relating CMT to birth trauma [8]. A congenitally shortened SCM is torn at birth with formation of a hematoma. Subsequent endomysial fibrosis results in deposition of collagen and migration of fibroblasts around individual muscle fibers that undergo atrophy. This theory is supported by findings from Cheng et al. [9] and Hollier et al. [10] that note a history of difficult birth associated with 30–60 % of patients with CMT. In our case, the patient was born by caesarean section at 38 weeks gestational age due to breech presentation. Some hypotheses suggest that abnormal fetal presentation and resultant birth trauma were initially considered to be the cause of CMT. Such presentations include breech presentation and transverse lie presentation.

Birth trauma was historically believed to result in hematoma formation in the SCM, leading to fibrous contracture of the muscle [11]. However, the theory does not explain the occurrence of CMT after caesarean sections which urges other hypothesis as the patophysiology of CMT. Davids et al. [12] have postulated it as sequelae of an intra-uterine or perinatal compartment syndrome. Lee et al. [13] concluded that intra-uterine constraint appear to be associated with ultrasonographically detected severe fibrosis involving the entire SCM muscle in early presenting CMT.

Several authors have advocated surgical treatment after the age of 1 year to allow time for possible spontaneous recovery [14–16]. Others have reported that a better and faster recovery of craniofacial asymmetry was achieved by correcting torticollis at an early stage, probably only a few weeks [16,17]. The disadvantages of early surgical treatment include more frequent wound breakdown, formation of hematoma, and superficial wound infection. As a result, it was recommended that the ideal patient age for muscular torticollis surgery is between 1 and 4 years [18,19]. Some authors reported that complete restitution of normal craniofacial asymmetry can be expected in a child surgically treated at less than 5 to 6 years of age [19–21]. Literature stated that patients with older age, and consequently, with more severe deformity, may require a bipolar release both at sthe sternal and clavicular site, as well as at the muscle's insertion at the mastoid process [6]. There is also concern regarding recurrence deformity because the clavicular head may reattach to the clavicle head forming a lateral band that may require secondary release [6].

In our case, the patient underwent complete unipolar release and such surgical strategy managed to overcome the deformity. The completeness of the release was confirmed by releasing the fibrotic SCM until fat around carotid sheath was found. We preferred to perform a complete unipolar release instead of bipolar release because the concern of releasing the sternocleidomastoid muscle at its mastoid attachment, places both the facial and the spinal accessory nerve at risk. It was easier to identify fat around the carotid sheath with a pulsating rhythm beneath it. After the release, we also excise a small portion of fibrotic muscle (0,5 cm) at the distal insertion of sternal head and clavicular head to ensure that recurrence did not occur.

The patient eventually regained full cervical range of motion, and significant improvement of CMA angle (from 14 to 0 degree post-operatively) after undergoing unipolar sternocleidomastoid release and three months of aggressive physical therapy. The CMA was defined as the angle between a line connecting the lower margins of the mandibular angles and a line drawn along the upper border of the C7 vertebral body [22]. This support the findings from Lee et al [21] reported that patients between the ages of 6 and 16 years had good operative results [23]. We also found no complication during the follow-up period. Other authors reported that the upper limit for good results after surgery for muscular torticollis is 12 years old [15]. It has also been suggested that if surgical treatment is delayed until the patient is 12 to 14 years of age, complete correction of the facial asymmetry is impossible [20,24].

4. Conclusion

Congenital muscular torticollis is a rare condition that when left untreated can cause fibrosis of the cervical musculature with progressive limitation of head movement, eye movement disorder, craniofacial asymmetry, and compensatory asymmetrical spine curvature that worsens with age. In those children who fail conservative management or present after the first year of life, satisfactory results can only be obtained with surgical release of the sternocleidomastoid muscle. It is important to perform thorough surgical release until carotid sheath is within vicinity. This procedure is generally well-tolerated with no major complications. An intensive regimen of postoperative physical therapy is crucial in enhancing the benefit of surgery.

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Ethical approval

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Consent

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Author contribution

Aryadi Kurniawan contributes in writing the paper and performing the surgery.

Anissa Feby Canintika contributes to data collection and writing the paper.

Registration of research studies

Does not need any registration.

Guarantor

Aryadi Kurniawan is the sole guarantor of this submitted article.

Declaration of competing interest

The authors declare no conflicts of interest.

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