Bilateral congenital torticollis: a case report with 25 years of follow-up

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Unilateral congenital sternocleidomastoid muscle (SCM) contracture causing torticollis is well known. Although the unilateral muscular torticollis is quite often recognized, a bilateral contracture of SCM muscle is very rare. A review of the literature showed only three cases of bilateral congenital torticollis reported over the last two decades. We present a case report of a boy with congenital bilateral torticollis with 25 years of follow-up. Bilateral tenotomies of the right SCM were performed and the child was immobilized in Schanz's cervical orthosis. Three months after operative procedure, the physical examination indicated an increasing contracture of the left SCM with time. A similar operative procedure was applied to the left SCM. The follow-up examinations showed good wound healing and a positive outcome. Bilateral congenital

muscular torticollis is a very rare form of muscle skeletal disorder. We describe a surgical treatment of such deformation that ended with a satisfactory result confirmed through a 25-year follow-up. *J Pediatr Orthop B* 26:585–588 Copyright © 2017 The Author(s). Published by Wolters Kluwer Health, Inc.

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Introduction

Unilateral congenital muscular torticollis, also known as wry neck, is a common condition caused by shortening and fibrosis of the sternocleidomastoid muscle (SCM). This condition is usually discovered during the first few weeks of life. It is characterized by the tilt of the infant's head to one side, with difficulties turning to the opposite side. A nontender mass attached of the SCM at the side toward the head is tilting can also be noted.

The bilateral contracture of the SCM is a very rare condition. We present a case of bilateral muscular torticollis first diagnosed and operated in a child aged 12 and a 25-year follow-up.

Case report

A 12-year-old boy was referred to the orthopedic department from a general-doctor practice. He was a second child of a nonconsanguineous marriage from full-term vaginal delivery in breech position. There was no family history of congenital deformities. He showed no other abnormalities. That particular patient had not been treated by any doctor before. All data considering his health status were provided by his parents, who reported

Fig. 1

Frontal view – age 12.

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Fig. 2

Back view – age 12.

that first symptoms of torticollis appeared in a preschool age.

The physical examination of the child indicated asymmetry of the head and face and a smaller right cheek. The lines of the mouth and eyes were crossing on the right part of the face. The head presented an altered, compulsory position of bending to the right side with the chin rotated to the left by $\sim 20^\circ$ (Figs 1–3). The observed aggravated tension of both SCM widened the shape of the neck and restricted its movements. The difference in shortening on both SCM was 2 cm. Range of motion (ROM) in the neck was limited for rotation to 30° and for lateral flexion to 20° . ROM on the left side was better and was 40° for rotation and 30° for lateral flexion. Infants of 2 months of age had a median muscle function score of 1, which makes it difficult for the baby to turn the head side to side. Examination of the spine showed compensatory deformation in the cervical and upper thoracic part, an increased thoracic kyphosis, and elevation of the right shoulder. Radiographs did not show any structural deformation of the vertebrae (Figs 4 and 5). As a treatment, the upper and lower tenotomies of the right SCM were performed and the child was immobilized in Schanz's cervical orthosis. Three months after the operative procedure, the physical examination indicated an increasing contracture of the left SCM with time. The

Fig. 3

Lateral view – age 12.

head again presented an altered, compulsory position of bending now to the left side and the chin rotation to the right. A similar operative procedure was applied to the left SCM. The child was placed in cervical orthosis in maximal extension, without any rotation or head lateral bending. The follow-up examinations during a period of 6 months showed good wound healing and a positive outcome. The child and the parents were satisfied with the result of the treatment.

The patient was invited for re-examination 25 years after the initial treatment. The physical examination at the age of 37 years showed symmetrical rotation of the head and no restriction to flexion or to extension of the cervical spine (Figs 6 and 7). The ROM for rotation was to 90° on both sides. ROM for lateral flexion showed its highest range to about 60° both sides. The initial asymmetry of the face was hardly noticed. The patient is an agricultural physical worker. The previous treatment has no present effect on his well-being.

Fig. 4

Anteroposterio view – radiography.

Fig. 5

Lateral view – radiography.

Discussion

Congenital muscular torticollis, or wry neck, is described as the most common form of painless torticollis. First descriptions were reported by Cheselden in 1749 [1] and later by Anderson 1893 [2]. Unilateral SCM contracture causing torticollis is well known. However the etiology of CMT remains unknown. Possible causes include a lack of space *in utero*, resulting in local compartment syndrome or ischemia producing fibrotic SCM [3]. As the child's head is tilted toward the involved neck muscle and the chin rotated toward the contralateral shoulder, the diagnosis is made on physical examination at birth or shortly after [1,4]. The finding of a hardened mass of SCM muscle on the involved side facilitates diagnostics. The unilateral contracture is almost universally present after infancy and may be replaced later with a fibrous contracted band [5].

Many authors agree that the nonsurgical treatment with stretching and massage yields an excellent result in more than 90% of patients [1,6–10]. Others emphasize the fact that for such treatment to be very successful, it should be started within a few months after birth. If this or other treatments do not yield positive results, surgery can

Fig. 6

Frontal view – age 37.

sometimes correct the problem. Conservative management is rarely successful in patients presented in childhood [7,11]. Less than 10% of cases require a surgical approach if a significant restriction of motion (above 30°)

Fig. 7

Back view – age 37.

or facial asymmetry is present at school age [4,10,12]. Ferkel bipolar lengthening of SCM is a procedure of choice for those patients [13]. We are of the opinion that in the patients seen in childhood, surgical intervention should be considered the treatment of choice to avoid further irreversible changes.

Bilateral congenital muscular torticollis is a very rare form of congenital muscular torticollis. The medical literature is scarce on cases of bilateral torticollis, with just three cases described in Hungary in 1993 [14], in Singapore in 2009 [15], and in China in 2016 [16]. A birth injury or a defective embryogenesis could likely be a cause of unilateral condition. Other theories of the origin of bilateral torticollis include fibrosis of the SCM muscle, resulting from venous occlusion because of intrauterine persistent position. Finally, we should take into consideration a genetic, growth retardation, infectious myositis, or mix of a variety of factors caused. Torticollis is a well-known deformity; however, bilateral malformation might present a differential diagnostic problem to orthopedic surgeons as well as ophthalmologists and neurologists.

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The authors have contributed substantially to the submitted work, have made a significant contribution to the findings and methods in the paper and have reviewed and agree with the submission of the manuscript for review. All authors have read and approved the final draft.

Conflicts of interest

There are no conflicts of interest.

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