Rehabilitation Practice and Science

Volume 40
Issue 2 *Taiwan Journal of Physical Medicine and Rehabilitation (TJPMR)*

Article 6

12-31-2012

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Recommended Citation

Chou, Tzu-Yi; Yang, Pei-Yu; and Meng, Nai-Hsin (2012) "Congenital Thoracic Scoliosis Presenting as Torticollis: A casereport," *Rehabilitation Practice and Science*: Vol. 40: Iss. 2, Article 6.
DOI: https://doi.org/10.6315/2012.40(2)06
Available at: https://rps.researchcommons.org/journal/vol40/iss2/6

Case Report

Congenital Thoracic Scoliosis Presenting as Torticollis: A Case Report

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Torticollis is a manifestation of various underlying conditions that refers to a specific positioning of the neck, such that the head is tilted to one side with the chin turned toward the opposite direction. Torticollis can be classified into muscular and non-muscular types. Congenital thoracic scoliosis is a rare cause of non-muscular torticollis. We report the case of an eight-month-old female infant presenting with persistent torticollis at birth. The initial impression was muscular torticollis. However, her symptom persisted after rehabilitation. After a series of surveys, congenital thoracic scoliosis, a very rare cause of non-muscular torticollis, was diagnosed. Most cases of muscular torticollis can be managed with good results. Non-muscular causes of torticollis, such as congenital vertebral anomalies, should be considered if torticollis is found with no evidence of a muscular origin or if the response to a rehabilitation program is poor. (Tw J Phys Med Rehabil 2012; 40(2): 97 - 102)

Key Words: torticollis, congenital scoliosis

INTRODUCTION

Torticollis is a manifestation of a variety of underlying conditions. Congenital muscular torticollis is the third most prevalent congenital musculoskeletal abnormality in newborns.[1] The common causes of non-muscular torticollis are central nervous system lesions, ocular torticollis, unilateral atlanto-occipital fusion, brachial plexus injury, central nervous system lesion, Klippel–Feil Syndrome, Sandifer's syndrome, and benign paroxysmal torticollis.[2] Most cases of muscular torticollis can be treated with good results.[2] Non-muscular causes of torticollis should be considered if no evidence of muscular origin is found or if the response to a rehabilitation program is poor.

Scoliosis-induced torticollis occurs most often at the cervical spine. Cervical spine radiographic examination is suggested for the evaluation of non-muscular torticollis.[3,4] Congenital thoracic scoliosis, as a cause of non-muscular torticollis, is very rare. We report a case of congenital thoracic scoliosis that presented as torticollis and highlight the importance of the examination of whole spine, not only the cervical spine, in the evaluation of torticollis.

CASE REPORT

A female infant with a history of congenital ventricular septal defect was noted to have wry neck (head tilted to the left) at birth. There was no history of trauma

Submitted date: 16 December 2011 Revised date: 17 April 2012 Accepted date: 26 April 2012
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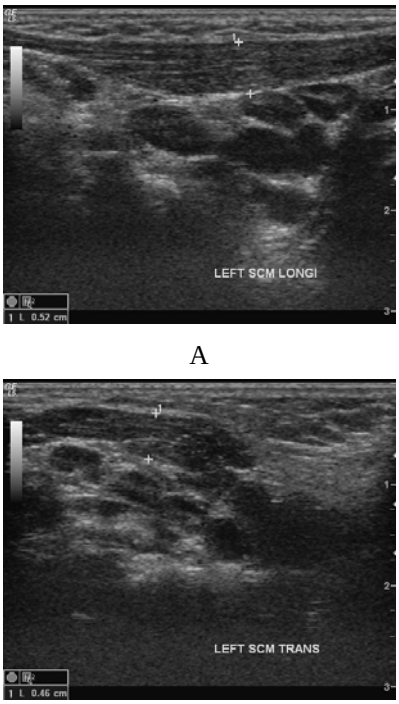
or frequent vomiting, difficult labor, breech deliveries, or caesarean section. Muscular torticollis involving the left sternocleidomastoid (SCM) muscle was diagnosed. The infant underwent a rehabilitation program that included positioning and stretching of the left SCM at a local hospital for six months, but symptoms persisted. She was brought to our clinic for persistent torticollis at the age of eight months. Head tilting to left and turning to right, as well as mild facial asymmetry, were noted. No signs of infection were evident, and low hairline, ptosis, or any palpable mass over the left SCM muscle existed. Her weight and height were appropriate for her age. The range of motion of her neck was not limited. No associated conditions, such as clubfoot, developmental hip dysplasia, and congenital metatarsus varus, were found.

Soft tissue ultrasonography was performed to evaluate the muscular torticollis, which showed no hyperplasia or fibrotic tissue over the left SCM (Figure 1A and 1B). She was referred to an ophthalmologist to evaluate strabismus. After examination, no strabismus was found. Spinal X-ray showed C-shaped kyphoscoliosis of the thoracolumbar spine (T3–L1), with convexity toward right side, an apex at T7, and a Cobb's angle of 34° (Figure 2A and 2B). Thoracic–lumbar spine CT was performed, which showed scoliosis of the thoracic spine attributed to multiple vertebral segmentation failure at the T1–T8 level (Figure 3A and 3B).

Other congenital malformations are commonly seen in congenital scoliosis, such as cardiac abnormalities (10%), genito-urinary abnormalities (25%), and intraspinal anomalies (40%).[5] In the current case, the patient also had congenital heart disease and a perimembranous type ventricular septal defect. The ventricular septal defect was diagnosed with cardiac sonography at one month of age. The defect closed spontaneously by aneurysmal transformation at the age of five months. After a series of studies, she was referred to an orthopedic surgeon for further management. Conservative treatment and follow-up on the progression of the scoliotic curve was suggested by the orthopedic surgeon. The orthopedic surgeon also suggested surgical treatment if the scoliotic curve progresses.

DISCUSSION



A

B

Figure 1. (A)Longitudinal view (B)Transverse view of left sternocleidomastoid muscle. Ultrasonographic images reveal no abnormal echogenicity nor increased thickness in the left sternocleidomastoid muscle (SCM).

(A) (B)

Figure 2. (A) Anteroposterior view (B)Lateral view of thoracolumbar spine. Spinal X-ray showed C-shaped kyphoscoliosis of the thoracolumbar spine (T3–L1).

(A)

(B)

Figure 3. (A)Anterior view(B)Posterior view of CT, thoracolumbar spine without contrast, after 3D reconstruction: showing multiple vertebral segmentation failure at T1-8 level and scoliosis of thoracic spine.

Most cases of torticollis discovered at or near the time of birth present with congenital muscular torticollis. The incidence of congenital muscular torticollis throughout the newborn period can be as high as 4/1000.[2] However, up to 80% of the cases improve by the time the child reaches one year with conservative treatment. Conservative treatment involves passive stretching of the involved SCM, with the shoulders stabilized and the head rotated to the side of the involved SCM and laterally tilted to the opposite side. Surgery is usually performed after the child reaches one year if persistent loss of range of motion, failure of conservative treatment, or progressive craniofacial asymmetry are present.[6,7]

Most cases of muscular torticollis can be treated with good results. Non -muscular causes of torticollis, such as congenital vertebral anomalies, should be considered if no evidence of muscular origin torticollis is found or if response to a rehabilitation program spanning more than six months is poor. In our case, the soft tissue echo showed no obvious muscular abnormality in the SCM, and the response to physical therapy was poor. Evaluation of the cause of non-muscular torticollis should be considered in this situation. A thorough history taking and physical examination are required. Non-muscular torticollis has various etiologies. A study involving 53 patients showed that the prevalence of non-muscular causes of torticollis in children could be as high as 18.4%. The most frequent cause is underlying neurologic disorder (51%), Klippel–Feil anomalies come second (30.2%), and clavicle fractures (3.8%), C1-2 rotary subluxations (3.8%), inflammatory conditions (3.8%), unexplained condition (7.6%) follow.[3] Radiographic examination of the cervical spine should be arranged if the patient has no history of trauma and if typical findings associated with sternocleidomastoid tightness for identifying congenital scoliosis are not manifested.[3,4,8] In our case, the bony abnormality occurred at the thoracic level. This condition is quite rare, highlighting the need to address the abnormality of whole spine, not only that of the cervical spine, in the differential diagnosis of non-muscular torticollis.

Other causes of scoliosis include trauma, septic torticollis, tumor, and so on. Radiographic examination is suggested in traumatic related torticollis.[3] Septic torticollis results from otolaryngologic infection or spondylodiscitis. Chronic torticollis is congenital, postural, or malformative. Tumoral diseases must be considered for recurrent forms, for torticollis with neurological abnormalities,[9] or for patients with specific underlying diseases, such as spinal epidural hematoma related torticollis in hemophilic patients.[10]

Congenital scoliosis is defined as a lateral curvature of the spine that results from malformations of the vertebra. The etiology of congenital scoliosis remains unknown. Genetic and environmental factors, vitamin

deficiencies, and drugs have all been implicated in the development of vertebral abnormalities.[11] The malformations of the vertebra include failure of segmentation or formation, as well as a combination of these defects. Other congenital malformations, such as cardiac abnormalities (10%), genito-urinary abnormalities (25%), and intraspinal anomalies (40%), are commonly seen in congenital scoliosis.[5] Frequent clinical and radiographic follow-up is required in infants diagnosed to have a vertebral anomaly, even in the absence of prominent scoliosis in an X-ray.[12] A thorough preoperative evaluation is recommended before deciding on any treatment protocol. Understanding congenital syndromes and knowing how these may affect both spinal deformity and their interaction with the treatment are necessary, including how they may result in additional morbidity.[13]

The type and progression of scoliosis will affect treatment planning. If the thoracic cage is involved, expansion thoracostomy and vertical expandable prosthetic titanium rib are indicated.[14] The diagnosis of thoracic insufficiency syndrome associated with early fusion is currently promoting new surgical techniques intended to preserve more vertebral growth in young patients with progressive scoliosis. A child who has extensive thoracic fusions or one who had an operation for fusions involving the proximal thoracic spine is under the highest risk for restrictive pulmonary disease.[15]

Early detection of scoliosis can provide more information to aid in the selection of treatment. Anomalies of vertebral development may present at birth, but are difficult to detect immediately. The detection of the curvature is always delayed until the child begins to sit or stand. In the current case, the early sign of thoracic scoliosis presented as torticollis even before the curvature became apparent. Thus, non-muscular causes of torticollis, such as congenital vertebral anomalies, should be considered if no evidence of a muscular origin is found or if the response to the rehabilitation program is poor.

CONCLUSION

Congenital thoracic scoliosis is a rare cause of torticollis. Most cases of muscular torticollis can be managed with good or excellent results. In the case of torticollis, if no evidence of muscular origin torticollis exists or if the response to the rehabilitation program is poor, non-muscular causes of torticollis, such as congenital vertebral anomalies, including thoracic lesions, should be considered.[16]

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以斜頸爲表現的先天性胸椎側彎：病例報告

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斜頸(torticollis)是指外觀上經常保持頭頸部傾向一邊，而臉轉向另一邊的姿勢。造成斜頸的原因很多，可以分爲肌肉性或非肌肉性的成因，其中以肌肉性成因佔最多數。

本篇報告一位 8 個月大的女嬰，出生時就有斜頸的症狀，原先診斷爲肌肉性斜頸，經復健治療成效不佳，後來進一步檢查，發現是先天性胸椎側彎。先天性胸椎側彎患者的脊椎異常，在出生時就已存在，但側彎的彎度在出生時不易察覺，通常遲至患者學會坐立或站立時，脊椎的彎度才會被察覺，而本個案的胸椎側彎在出生時就以斜頸爲表現。骨發性斜頸通常是以頸椎的異常較常見，本個案因先天性胸椎側彎而造成斜頸較罕見。一般而言，肌肉性斜頸經復健治療的成效大多十分顯著，如果發現斜頸的個案並沒有明顯的胸鎖乳突肌異常，或對復健治療的效果不顯著時，應將非肌肉性的成因，例如脊椎異常，列入斜頸的鑑別診斷。（台灣復健醫誌 2012；40(2)：97 - 102）

關鍵詞：斜頸症（torticollis），先天性脊柱側彎（congenital scoliosis）

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