Torcicolo Muscular Congénito: Um Diagnóstico na Idade Adulta

Congenital Muscular Torticollis: A Diagnosis in Adult Age

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Resumo

Paciente do sexo feminino, 21 anos, encaminhada por dor e leve limitação funcional do ombro esquerdo. Houve edema da banda fibrosa da cabeça clavicular do músculo esternocleidomastoideo esquerdo. O estudo imagiológico revelou aumento da metade interna da clavícula esquerda e espessamento difuso da cabeça clavicular do músculo esternocleidomastoideo esquerdo, sugerindo fibrose. Após a cirurgia, outro programa de reabilitação foi iniciado e a paciente teve uma boa recuperação.

Palavras-chave: Adulto; Músculos do Pescoço; Torcicolo/congénito; Torcicolo/reabilitação

Abstract

A 21 years old female patient was referred due to pain and slight functional limitation of the left shoulder. There was fibrous band swelling of the left sternocleidomastoid muscle's clavicular head. The imaging study revealed enlargement of the inner half of the left clavicle and diffuse thickening of the clavicular head of the left sternocleidomastoid muscle, suggesting fibrosis. After surgery, another rehabilitation program was initiated and the patient had a good recovery.

Keywords: *Adult; Neck Muscles; Torticollis/congenital; Torticollis/rehabilitation*

Introduction

Congenital muscular torticollis (CMT) is the third most frequent cause of congenital musculoskeletal anomaly and consists of neck deformity involving primarily a shortening of the SCM muscle that is detected at birth or, usually, soon after birth.[1] Early diagnosis and treatment, as well as active parental involvement are critical to therapeutic success.[2] In light with the literature, the situation foresaw poor prognosis.[2] Rehabilitation treatment is not only important as the first therapeutic approach, but also as a post-surgical treatment.[3] The present case report aims to describe a case of CMT, diagnosed in an adult patient.

Case Report

A 21 years old female patient was observed by Physical Medicine and Rehabilitation (PMR) due to left shoulder pain with mechanical rhythm, with several years of evolution. It had metameric projection to the base of the cervical spine, with an insidious installment, scoring 5/10 on the pain numerical scale. Neuropathic implication was ruled out and there were no motor deficits associated. She also presented dorsal and lumbar pain for long periods (>30 minutes) of orthostatism.

When asked about her neck deformity, the patient mentioned not to have acknowledge it until six years ago. Since, she was consulted by an orthopedic surgeon

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(10/12/2012), who recommended swimming and the use of an insole (which she never used). In childhood, she had recurrent otitis and pharyngitis, she had a tonsillectomy and sinus surgery at 7 years old. Her family history was irrelevant. Her mother's pregnancy was followed in a German hospital, uneventfully. She was born at 38 weeks, discotic delivery, cephalic presentation, using forceps and suction cups; her mother's serologies were negatives. She was born with 51 cm of height and weighed 3.450 kg. No known history of hereditary diseases.

At our observation, her lingered clinical condition consisted of a nodular fibrous band of the left SCM's clavicular head, with no pain evoked by palpation (Fig. 1), cervical posture tilted to the left and right cervical rotation (Fig. 2), active and passive limitations of the right lateral flexion (20º), the left rotation (70º) and the cervical extension (30º) (Fig. 3). Patient present also a left gibbosity, winged scapula (Fig. 4), disability of the scapulohumeral rhythm (Fig. 5), as well as limitation in the active (140º) and passive (160º) range of motion in both lateral and anterior elevation of the left shoulder. This was probably caused by subacromial conflict, secondary to antero-lateral scapula translation and left clavicle elevation (Fig. 6 – without evident deformity or associated crackling), which conditioned the elevation of the entire left shoulder girdle. Hawkins Kennedy test and Yocum's test were positive (on the left shoulder).

Figure 1 - Nodular fibrous band of left SCM's clavicular head.

Figure 2 - Left cervical tilt and right cervical rotation posture.

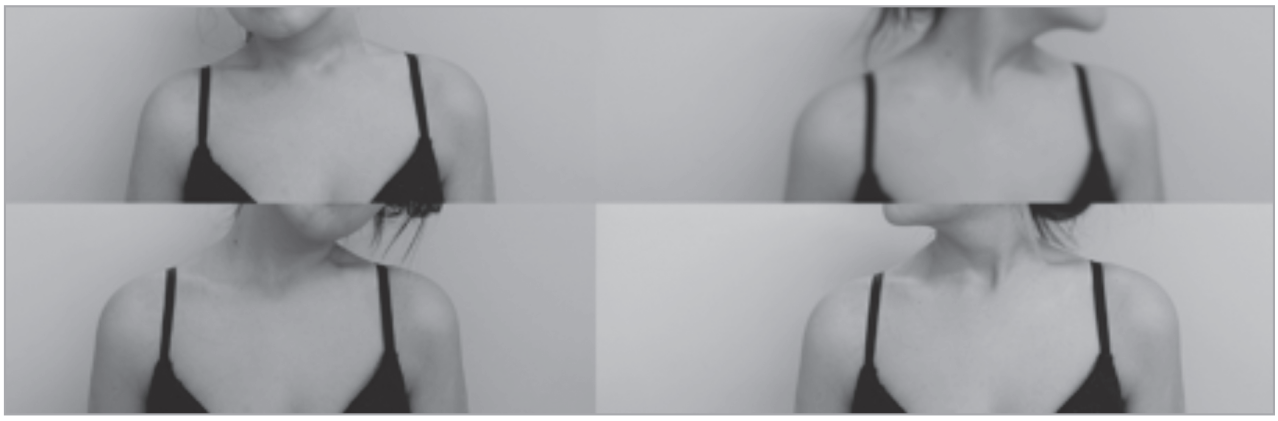

Figure 3 - Limitation of active range of motion of the cervical area.

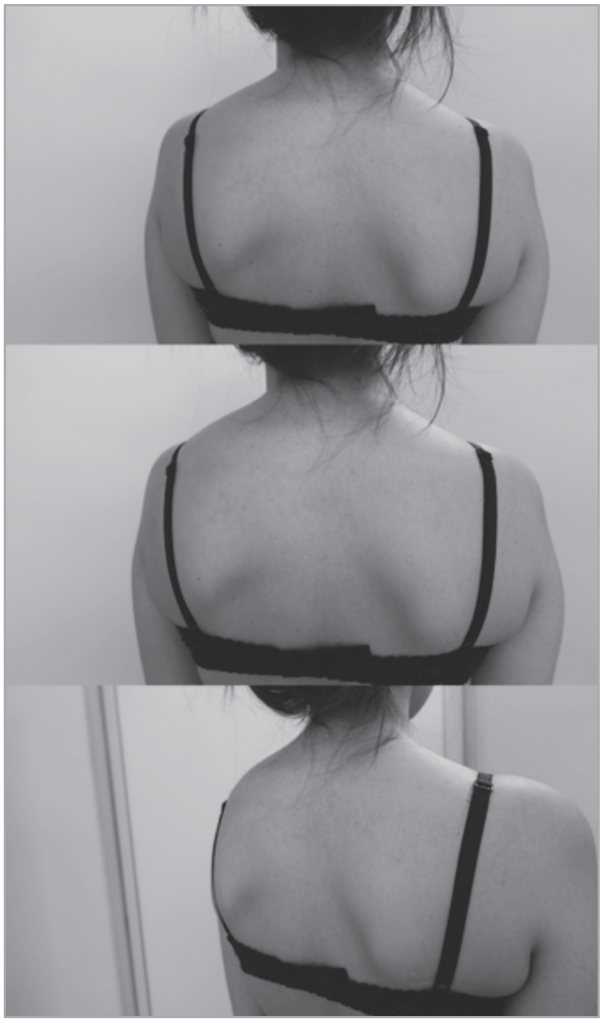

Figure 4 - Left winged scapula.

Figure 5 - Left alteration of the scapulohumeral rhythm.

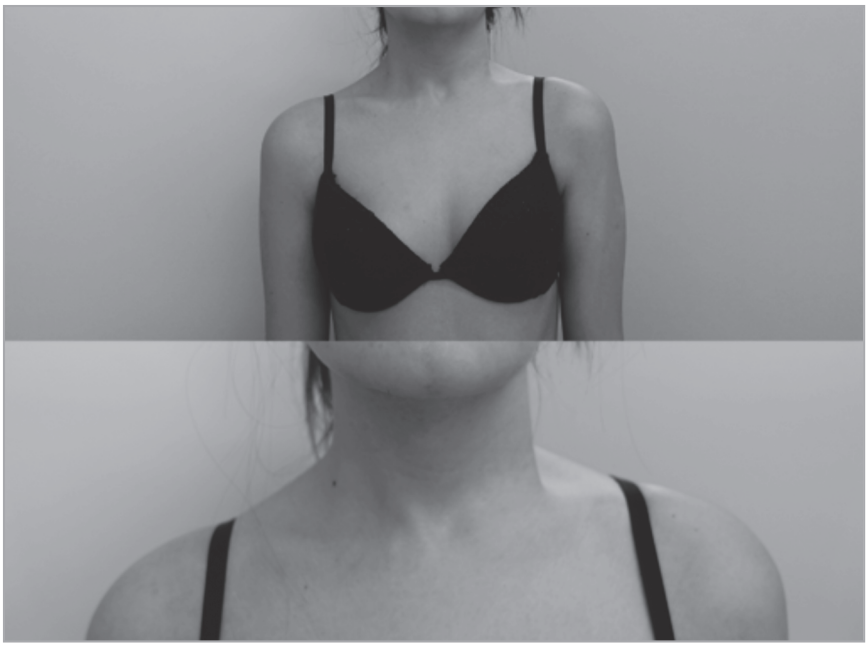

Figure 6 - Left clavicle elevation.

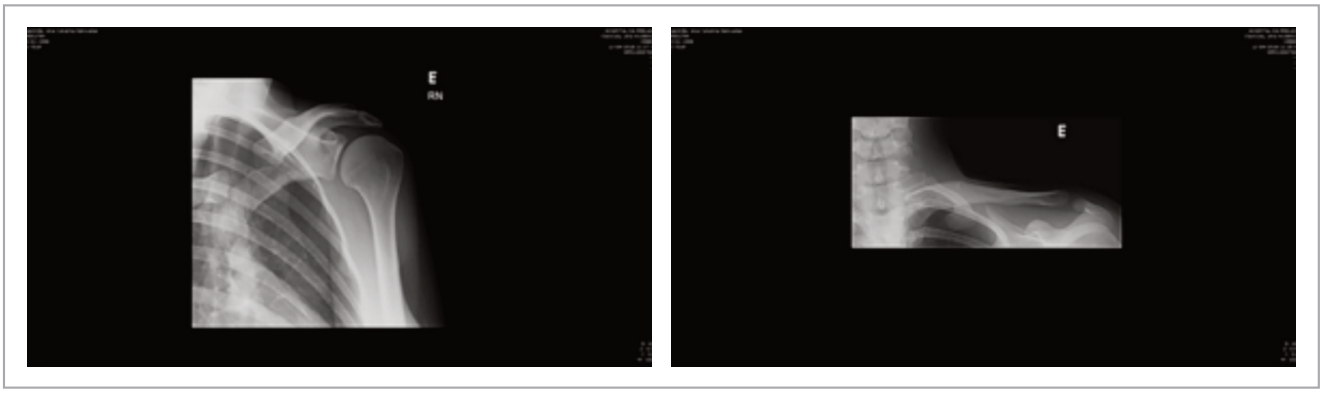

Figure 7 - Left shoulder radiography.

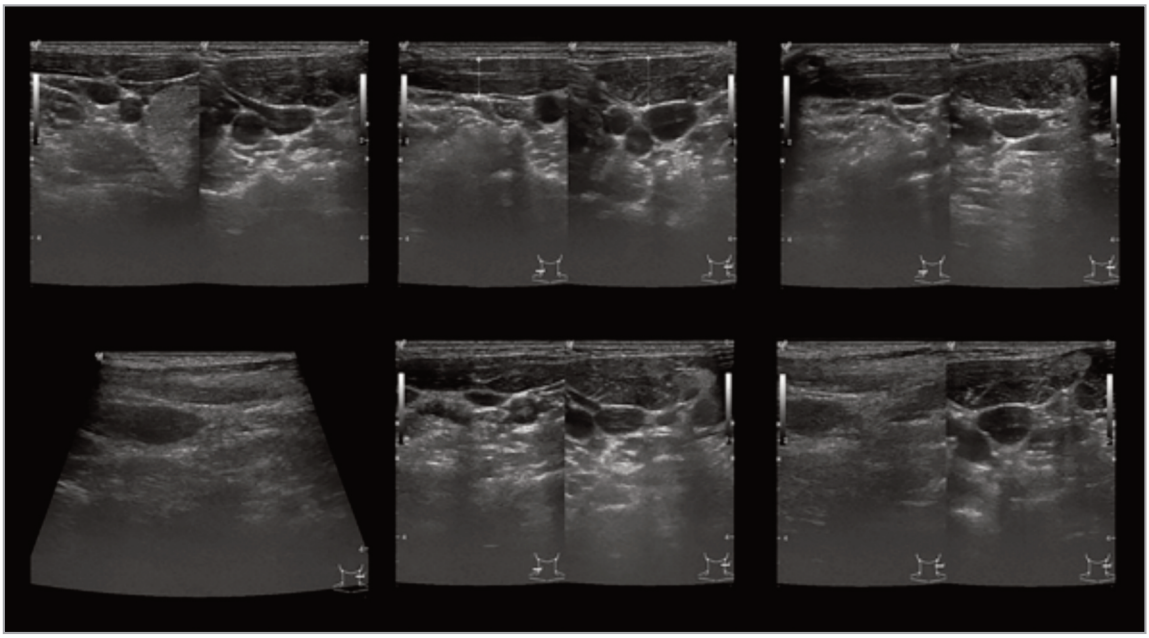

Figure 8 - Neck's soft tissue ultrasound.

Remaining musculoskeletal and neurological examinations revealed to be normal.
The patient underwent imaging study (extra-long radiography of the lower limbs and spine) which revealed (22-01-2018): "Lower limbs of equivalent dimensions (79.5 cm to the left and 79.4 cm to the right). Slight dorso-lumbar scoliosis". Unfortunately, it was not possible to observe the images on this exam, so the magnitude of scoliosis has not been evaluated. Left shoulder radiography showed (12-04-2018): "Enlargement of the inner half of the left clavicle (possible bone callus resulting from an old fracture)" (Fig. 7).

Neck's soft tissue ultrasound (12-04-2018): "Diffuse thickening of the clavicular head of the left SCM muscle, with clear asymmetry compared to the contralateral. There is some heterogeneity and hyperechogenicity of the tendon bundle, suggesting fibrosis, although the findings should be clinically correlated" (Fig. 8).

Posterior observation of childhood's photographs allowed to verify the existence of cervical posture in left inclination and right rotation, as well as tension on the left SCM.

CMT and traumatic rupture of the SCM with associated fibrosis emerged as the main diagnostic hypotheses. As secondary diagnostic hypotheses, we considered Sprengel's deformity, a rare congenital scapular deformity, which can also be associated with alterations in the static position of the head, scoliosis and clavicle deformity. However, our examination did not show deformity or elevated position of the scapula, cranial asymmetries or in other bone structures, hypoplasia of the neck and shoulder muscles nor passive limitation of the left shoulder joint amplitudes. Additionally, the radiological study performed did not reveal any bone abnormalities, like fusion of cervical vertebrae (Klippel-Feil syndrome, with an association of 7% to 42%), therefore, this hypothesis was eventually discarded and it was decided not to perform any additional imaging

study of the scapula, such as a computed tomography. The fact that the patient did not have any motor deficit against high resistance at the level of the left SCM and trapezius muscle, ruled out the possibility of accessory nerve injury (injury to the cranial nerve XI), thus, electromyography was not requested.

A subsequent medical appointment with the joint assessment of an experienced orthopedic surgeon, well versed in pediatric pathology, was an important contribution to the exclusion of these last three diagnostic hypotheses.

After analyzing the results of the complementary diagnostic exams and their clinical correlation, it was assumed a CMT undiagnosed in childhood was the most likely cause for the pain and biomechanical changes. There are many scientific studies with clear evidence that changes in neck muscles (such as the SCM muscle) can cause positional changes in the scapula and scapulohumeral dyskinesia, and, consequently, static postural changes and vertebral scoliosis. Muscle imbalance is usually the result of repetitive wrong movements and sustained postures in cases such as this, which cause a mechanical overload leading to increased tension and shortening of certain muscle groups (such as agonists of a given movement) and weakening and inhibiting of others (their antagonists). Hence, an alteration in biomechanics of the neck muscles, such as an increased tension in the SCM and cervical extensors muscles (levator scapulae, upper trapezius, suboccipitals), or of the pectoralis muscles (more rarely in the biceps, brachialis and brachioradialis) can cause inhibition of the scapular stabilizer muscles (rhomboideus, middle and lower trapezius and anterior serratus), as well of the cervical flexors (more rarely, shoulder extensors) - upper crossed syndrome (also known as proximal crossed syndrome or shoulder crossed syndrome).[4-8]

Rehabilitation program was initiated aiming to functional gain and improvement of associated symptoms; however, it was unsuccessful. It was proposed surgical releasing of the left SCM's clavicular insertion, that she realized at 20/11/2018. After surgery, another rehabilitation program was initiated, with shoulder's articular kinesiotherapy, muscle strengthening exercises focusing on the humeral head depressor muscles, scapula stabilizers and external rotators, focused on functional tasks and right scapula's position. Neuro-muscular electrical stimulation of the posterior deltoid fibers, muscle relaxation massage of the cervico-scapular waist and stretching exercises of the anterior chains were performed. This program also included home exercises teaching.

Follow-up

The clinical condition had a favorable evolution, with progressive functional recovery since the beginning of the rehabilitation program. At 12 weeks of physiatric treatment after surgery, partial recovery of active cervical joint amplitudes was seen (right lateral flexion 35º, left rotation 80º and cervical extension 45º), and also total recovery of active left shoulder's amplitudes, however, keeping a slight change in the scapulohumeral rhythm (Fig. 9).

One year after surgery, the patient's state is overlapping with the last assessment, described above. Since she is a young woman, it is also worth highlighting and valuing the aesthetic damage that is likely to be permanent.

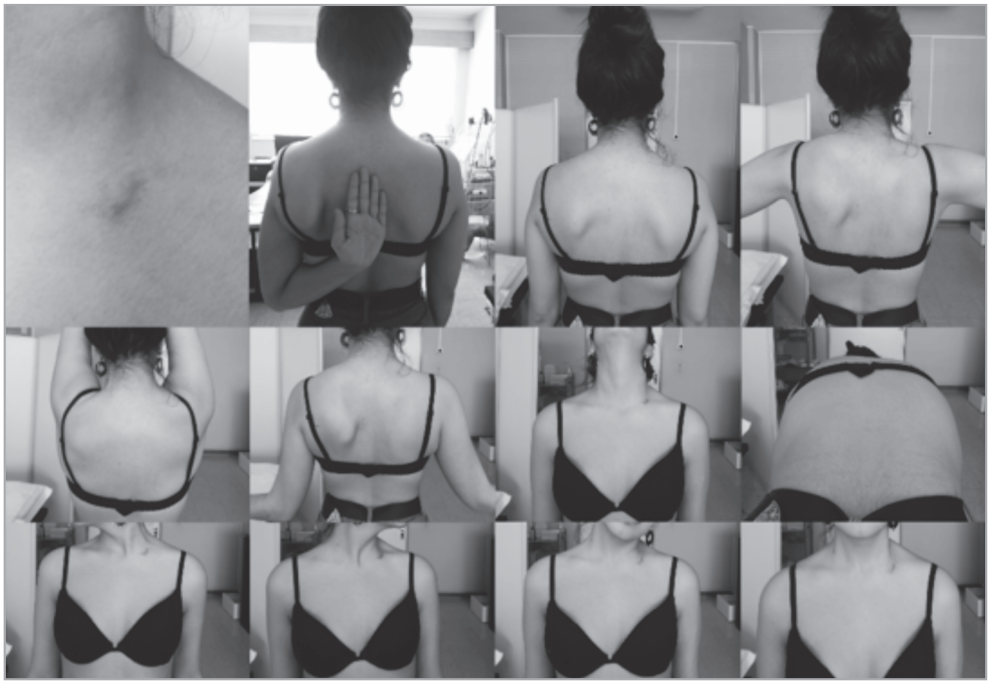

Figure 9 - Twelve weeks after surgery.

Discussion

Torticollis consists on a deformity, congenital or acquired, which leads to lateral inclination towards the homolateral shoulder with head rotation towards the other side. CMT, which involves a shortening of the SCM muscle, is the third most frequent cause of congenital musculoskeletal anomaly and it is detected, usually, soon after birth.[9] Its incidence varies from 0.3% to 1.9% of newborns.[10] Although there are many theories about its etiology, the exact pathogenesis remains unknown.[1] An association between the presence of CMT and craniofacial dysmorphias (with plagiocephaly being the most frequent) and hip's development dysplasia (with an incidence up to 20%) has been described.[11]
This case report refers to CMT with nodular fibrosis. Exhaustive clinical history and a complete physical examination are fundamental to exclude other differential diagnosis.[12] Complementary, ultrasound is the standard imaging modality to evaluate CMT.[13]

The present description highlights the importance of early diagnosis and treatment as well as active parental involvement are critical to therapeutic success. Indeed, as previous reported in the literature, infants with CMT treated early, either at home or in the outpatient clinic, showed completely recovered normal neck movement in a short time.[12,14] In the described case, as result of the late diagnosis, the patient presented several factors of poor prognosis such as swelling of the SCM, limitation of passive neck rotation and late presentation age. These factors, as consequence, compromised the rehabilitation outcomes, which lead to surgical procedure.
The present case also stresses the relevance of the investigation of present and past history. In fact, the previous related history, namely childhood's photographs, raised the diagnostic hypothesis of sequelae of a previously undiagnosed CMT. Therefore, the diagnostic suspicion is essential to achieve full resolution of CMT symptoms to exclude the minimal risk of relapse.

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Referências / *References*

1. Sargent B, Kaplan SL, Coulter C, Baker C. Congenital Muscular Torticollis: Bridging the Gap Between Research and Clinical Practice. Pediatrics. 2019;144:e20190582. doi: 10.1542/peds.2019-0582.
2. Tessmer A, Mooney P, Pelland L. A developmental perspective on congenital muscular torticollis: a critical appraisal of the evidence. Pediatr Phys Ther. 2010 ;22:378-83. doi: 10.1097/PEP.0b013e3181f940f3.
3. Kaplan SL, Coulter C, Sargent B. Physical Therapy Management of Congenital Muscular Torticollis: A 2018 Evidence-Based Clinical Practice Guideline From the APTA Academy of Pediatric Physical Therapy. Pediatr Phys Ther. 2018;30: 240-90.
4. McQuade K, Dawson J,Smidt GL. Scapulothoracic muscle fatigue associated with alterations in scapulohumeral rhythm kinematics during maximum resistive shoulder elevation. J Orthop Sports Phys Ther. 1998;28: 74-80.
5. Song SJ. Effect of Craniocervical Flexion on Muscle Activities of Scapula Upward Rotator Muscle During Push-Up Plus Exercise in Subject With Winging of Scapula. Phys Ther Korea. 2014; 21:48-52.
6. Kim EK, Kim JS. Correlation between rounded shoulder posture, neck disability indices, and degree of forward head posture. J Phys Ther Sci. 2016;28:2929-32. doi: 10.1589/jpts.28.2929.
7. Mujawar JC, Sagar JH. Prevalence of Upper Cross Syndrome in Laundry Workers. Indian J Occup Environ Med. 2019;23: 54-6.
8. Bayattork M, Seidi F, Minoonejad H, Andersen LL, Page P. The effectiveness of a comprehensive corrective exercises program and subsequent detraining on alignment, muscle activation, and movement pattern in men with upper crossed syndrome: protocol for a parallel-group randomized controlled trial. Trials. 2020;21: 255. doi: 10.1186/s13063-020-4159-9.
9. Cheng JC, Tang SP, Chen TM, Wong MW, Wong EM. The clinical presentation and outcome of treatment of congenital muscular torticollis in infants--a study of 1,086 cases. J Pediatr Surg. 2000;35:1091-6. doi: 10.1053/jpsu.2000.7833.
10. Kuo AA, Tritasavit S, Graham JM Jr. Congenital muscular torticollis and positional plagiocephaly. Pediatr Rev. 2014;35:79-87; quiz 87. doi: 10.1542/pir.35-2-79. Erratum in: Pediatr Rev. 2014;35:137.
11. Do TT. Congenital muscular torticollis: current concepts and review of treatment. Curr Opin Pediatr. 2006;18:26-9.
12. Carenzio G, Carlisi E, Morani I, Tinelli C, Barak M, Bejor M, Dalla Toffola E. Early rehabilitation treatment in newborns with congenital muscular torticollis. Eur J Phys Rehabil Med. 2015;51: 539-45.
13. Dudkiewicz I, Ganel A, Blankstein A. Congenital muscular torticollis in infants: ultrasound-assisted diagnosis and evaluation. J Pediatr Orthop. 2005;25:812-4. doi: 10.1097/01.bpo.0000184648.81109.75.
14. Kaplan SL, Coulter C, Fetters L. Physical therapy management of congenital muscular torticollis: an evidence-based clinical practice guideline: from the Section on Pediatrics of the American Physical Therapy Association. Pediatr Phys Ther. 2013;25:348-94. doi: 10.1097/PEP.0b013e3182a778d2.