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(CASE REPORT)

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Congenital muscular torticollis in young adultpatient: A neglected case.The efficiency of bipolar release

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Abstract

Congenital muscular torticollis (CMT) is a common postural deformity evident shortly after birth characterized by ipsilateral cervical lateral flexion and contralateral cervical rotation due to unilateral shortening of the sternocleidomastoid muscle. Untreated, the sequelae include facial asymmetry, tight bands, ophthalmic abnormalities, and altered neck posture.

To report a neglected CMT in 17-year-old female treated surgically by bipolar muscle release technic. The surgery was made via a double incision above the clavicular and mastoid with the liberation of the fibrous bands followed by intensive physiotherapy program. The patient was very satisfied after management.

Neglected CMT adult's patients can benefit from surgical treatment. Bipolar release is an adequate and complication-free method with postoperative physiotherapy program.

Keywords: Neglected congenital muscular torticollis; Cervical scoliosis; Surgery; Bipolar release technic; Physiotherapy

1. Introduction

Congenital muscular torticollis (CMT) is characterized by shortening and fibrosis of the sternocleidomastoid (SCM) muscle detected at birth or shortly after birth. This leads to a lateral inclination of the head to the ipsilateral shoulder and chin deviation to the opposite side [1, 2, 3].

Such affected patients need treatment to avoid torticollis-like complications. The twisted neck position can result in positional plagiocephaly and facial asymmetry if neglected [3].

A case of a neglected case of CMT in adulthood complicated by cervical scoliosis and facial asymmetry is reported.

2. Case report

17-year-old female without particular pathological antecedent was admitted to the neurosurgery department for congenital torticollis with neck pain and limited movement of the neck. This deviation has become painful for 3 years.

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The examination found a patient in good general condition. There was a limited range of motion upon rotation of the neck to the side. On examination, we see unilateral torticollis with the finding of a visible and palpable bridle in the right SCM region. On physical exam, a localized band of tissue with tethering could be palpated within the superior portion of the remaining SCM with rotation to the left. There was a facial asymmetry with hypertrophy of the left hemi-face (Figure 1). Neurological examination was normal. Other somatic examinations were normal. Laboratory investigations were unremarkable.

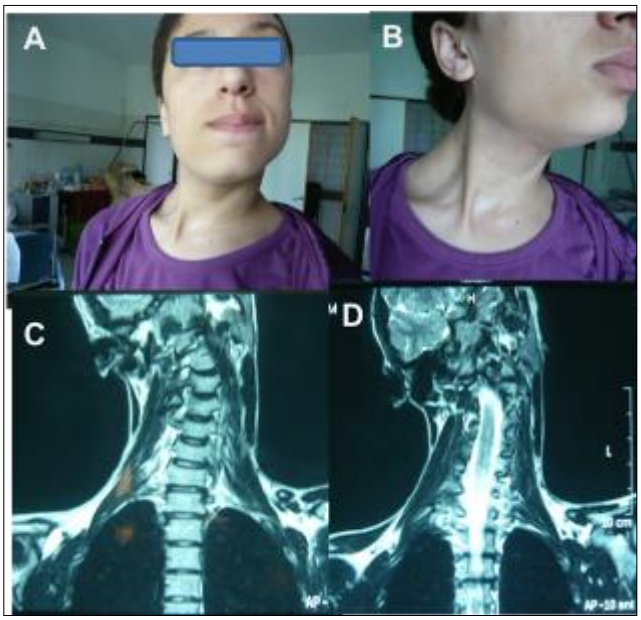

Figure 1(A, B) Pre-operative photo of the patient: note the cervical deviation with scoliosis and the retractile band of the right SCM muscle and Facial asymmetry. (C, D): Cervical MRI in coronal view in T1-W and T2-W images. Neglected congenital muscular torticollis in young adult

The cervical Magnetic resonance imaging (MRI) shows lateral deviation with cervical scoliosis and asymmetry of the two SCM muscles. The right SCM muscle showed a hyposignal in T1-W and T2-W MRI (Figure 1).

Surgical treatment was performed under general anesthesia using retroauricular and supraclavicular approaches (Figure 2). The right SCM muscle was released from its insertions in the mastoid process and clavicle. A fibrous bundle segment of the muscle was resected. Correction of the neck movement restriction was checked and tested intraoperatively. Finally, a bipolar release was made.

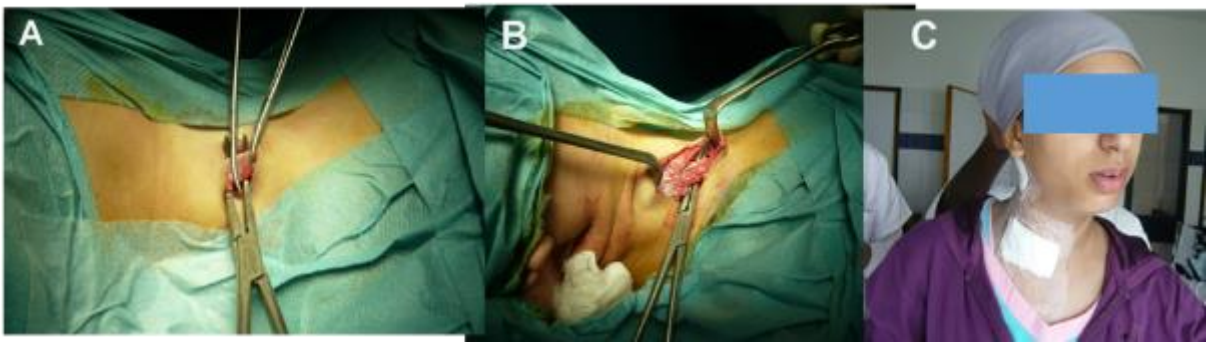

Figure 2(A, B): Per-operative view of inferior supraclavicular incision performing muscular release. (C): immediate postoperative photo of the patient: note the postoperative release of cervical deviation with less pain

The postoperative was favorable without complication. On awakening the patient felt well, the pain has subsided and the deviation is less obvious. Immobilization with a cervical brace was applied the entire day during the first 2 weeks. Three months of the rehabilitation program of manual stretching and active and passive exercises was instituted after brace removal.

Discussion

Congenital muscular torticollis (CMT), is recognized as unilateral contracture and shortening of the SCM muscle due to muscle atrophy and interstitial fibrosis, causing ipsilateral head tilt and turn. Its frequency in the newborn ranges from 0.3% to 2% [4, 5, 6].

Its etiology remains unclear, different theories have been proposed. The suggested causes include trauma during birth, ischemia, constitutional and growth arrest causes, compartment syndrome, hereditary predisposition, and neurogenesis.

Neglected or undiagnosed CMT in adults is quite rare, although it is the third most common congenital deformity in newborns (1). When left untreated at an early age, deficits in lateral and rotational range of motion can occur along with irreversible facial and skeletal deformities that develop over time [7]. Subtle cases can go unnoticed until early adulthood, with predominant fibrotic replacement in the SCM making physical therapy and chemodenervation mostly ineffective. Untreated, the sequelae include facial asymmetry, tight bands, ophthalmic abnormalities, and altered neck posture. The CMT in the adult are very rare but reported in the literature. It has been reported that CMT may result in secondary scoliosis over long-term follow-ups [8, 9, 10].

The diagnosis is usually performed by physical examination. CMT is usually recognized in neonates as a circumscribed palpable mass confined to the SCM, affected unilaterally, which may gradually disappear between 4 and 8 months of age or be associated with other orthopedic abnormalities such as hip dysplasia or lower extremity abnormalities.

Typical findings are a palpable indurated mass in the SCM muscle, with shortening of this muscle, which leads to the lateral inclination of the head to the ipsilateral shoulder and chin deviation to the opposite side. Asymmetrical development of the face is possible in adults.

MRI can show muscle atrophy and interstitial fibrosis and complication in neglected cases. Due to the presence of fibrous tissue, MRI characteristics include reduced mass signal strength on T2-W images relative to gradient recalled T1-W images [1,6,8].

Besides CMT, other causes of pseudodystonic torticollis at large include focal myopathies, rotational atlantoaxial subluxation, congenital Klippel–Feil anomaly, trochlear nerve palsy, and vestibular torticollis.

An accessory sternocleidomastoid can mimic a CMT and can result in torticollis because it causes a physical restriction preventing the neck from tilting and rotating to the opposite side[4]. In mild cases, the anomaly may remain undiagnosed until adulthood and can be confused with cervical dystonia and fibromatosiscolli.

If not treated, contracture of the SCM muscle may induce plagiocephaly or skull and facial asymmetry [1,7,8]. Once these problems have occurred, it is not possible to correct them after the period of growth potential has passed.

The mainstay of management of CMT is stretching exercise. About 90% or more of CMT cases are known to be cured with only stretching exercise and without any musculoskeletal complications.

Very few reports have been published on surgical treatment in neglected adult cases. Different surgical methods include SCM excision, the bipolar release of SCM with Z-plasty muscle bulk reconstruction, and tenotomy or release of the SCM [1]. The prognosis of children diagnosed and treated when they are older than a year is worse. In our case bipolar release was effective and safe with a prolonged rehabilitation program. Surgical treatment of neglected CMT in adults can be difficult due to adjacent tissue contractures, adhesions, and extensive fibrosis [6]. Facial asymmetry is very difficult to resolve after 4 years of age. Although different investigators have determined that surgical treatment is of little value after 4 years of age.

Concerning surgical treatment in neglected adult cases, very few reports have been published. The bipolar release technic is a simple technic with good results which can prove effective in alleviating pain, improving function, and cosmesis [1].

Surgical treatment led to cosmetic and functional improvements and relieved pain originating from the muscle imbalance brought about by the long-standing deformity [9, 10].

Consistent with the results found in the literature, is that bipolar release of both anterior and posterior muscle bellies, with resection if indicated due to extensive adhesions, is the best initial approach to treating neglected CMT in the adult population. An effective postoperative protocol of physical therapy is necessary.

3. Conclusion

Neglected congenital muscular torticollis adult's patients can benefit from surgical treatment. Bipolar release is an adequate and complication-free method with postoperative physiotherapy program.

Compliance with ethical standards

Disclosure of conflict of interest

The authors declare that there are no conflicts of interest.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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