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Thesis N° 313

Congenital Torticollis –wryneck– in children: surgical and post–operative management

THESIS

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ΒY

Ms. JAMMA Meriem

Born on May 22nd, 1999 in Marrakesh

TO OBTAIN A MEDICAL DOCTORATE

KEYWORDS

TORTICOLLIS- CONGENITAL- SURGERY- TENOTOMY- PHYSIOTHERAPY

JURY

| Mr. | R. EL FEZZAZI | | CHAIRPERSON |
|-----|--------------------------------|--------------|-------------|
| | Professor of Pediatric surgery | | |
| Mr. | A. EL KHASSOUI | | |
| | Professor of Pediatric surgery | | Supervisor |
| Mr. | EE. KAMILI |) | |
| | Professor of Pediatric surgery | | |
| Mr. | E. AGHOUTANE | <pre>}</pre> | JUDGES |
| | Professor of Pediatric surgery | J | |





Serment d'hippocrate

Au moment d'être admis à devenir membre de la profession médicale, je m'engage solennellement à consacrer ma vie au service de l'humanité.

Je traiterai mes maîtres avec le respect et la reconnaissance qui leur sont dus.

Je pratiquerai ma profession avec conscience et dignité. La santé de mes malades sera mon premier but. Je ne trahirai pas les secrets qui me seront confiés. Je maintiendrai par tous les moyens en mon pouvoir l'honneur et les nobles traditions de la profession médicale. Les médecins seront mes frères. Aucune considération de religion, de nationalité, de race, aucune considération politique et sociale, ne s'interposera entre mon devoir et mon patient. Je maintiendrai strictement le respect de la vie humaine dés sa

conception.

Même sous la menace, je n'userai pas mes connaissances médicales d'une façon contraire aux lois de l'humanité. Je m'y engage librement et sur mon honneur.

Déclaration Genève, 1948



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| 288 | AIT BELAID Wafae | Pr Ass | Chirurgie générale | |
| 289 | ZTATI Mohamed | Pr Ass | Cardiologie | |
| 290 | HAMOUCHE Nabil | Pr Ass | Néphrologie | |
| 291 | ELMARDOULI Mouhcine | Pr Ass | Chirurgie Cardio-vasculaire | |
| 292 | BENNIS Lamiae | Pr Ass | Anesthésie-réanimation | |
| 293 | BENDAOUD Layla | Pr Ass | Dermatologie | |
| 294 | HABBAB Adil | Pr Ass | Chirurgie générale | |
| 295 | CHATAR Achraf | Pr Ass | Urologie | |
| 296 | OUMGHAR Nezha | Pr Ass | Biophysique | |
| 297 | HOUMAID Hanane | Pr Ass | Gynécologie-obstétrique | |
| 298 | YOUSFI Jaouad | Pr Ass | Gériatrie | |
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| 323 | CHERKAOUI RHAZOUANI Oussama | Pr Ass | Neurologie | |
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| 325 | BENCHANNA Rachid | Pr Ass | Pneumo-phtisiologie | |
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| 327 | EL GHOUL Naoufal | Pr Ass | Traumato-orthopédie | |
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In Memory of My Grandparents ELhaí Moubarak et Lhaía Hadda, To the most generous and kind-hearted souls I have ever known, my beloved grandparents, Your lives were a testament to the true meaning of generosity and kindness. Through your actions, you showed me and everyone around you what it means to give without expecting anything in return, to care deeply, and to love unconditionally. You welcomed everyone with open arms and warm hearts, always ready to lend a helping hand or offer a comforting word. Your home was a haven of love, where laughter was shared and memories were made. Your kindness knew no bounds, and your generosity was felt by all who had the privilege of knowing you. The lessons you taught me through your selfless acts will stay with me forever. You taught me that true wealth is measured not by what we have, but by what we give. You showed me that kindness can change the world, one small act at a time. Thank you for the countless ways you touched my life and the lives of so many others. Your legacy of generosity and kindness will live on in our hearts and through the lives we lead.

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List of abbreviations:

| СМТ | : congenital muscular torticollis |
|---------|-----------------------------------|
| SCM | : sterno-cleido-mastoid. |
| DDH | : Dyplasia of the hip. |
| CT-scan | : computed tomography scan. |
| MRI | : Magnetic resonance imaging. |
| ENTs | : ear-nose-throat specialists. |



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The term torticollis comes from the Latin words tortus, meaning "twisted," and collum, meaning "neck." In 1912, it was defined as a deformity characterized by the head tilting to one side, with the neck twisted and the face deviated. Congenital muscular torticollis (CMT) is a painless condition that occurs in infancy, caused by the shortening of the sternocleidomastoid muscle (SCM) on one side. This leads to tilting of the head towards the affected side and rotation of the face and chin towards the opposite side(1). It is observed in 0.3–1.9% of all live births, (2)other studies indicate a ratio of 1 per 250 newborns being the third congenital orthopedic anomaly, more frequently following congenital hip dysplasia and clubfoot. (3)

There is evidence from certain authors indicating that torticollis often occurs alongside hip dysplasia.(4)

CMT is a condition commonly diagnosed at or right after birth. The shortening of the SCM can be associated to a fibrotic mass. the retraction of the muscle can be observed clinically or through an ultrasound examination. This can happen either during the development of the fetus or as a result of trauma during the perinatal period.(5)

The etiologies of CMT are not very well understood. In the past, it has been linked to birth trauma, prenatal or perinatal compartment syndrome, and limitations of the development of the sternocleidomastoid muscle due to intrauterine constraint. However, recent studies using immunohistochemical and gene expression techniques offer more compelling evidence for an impaired development of the SCM muscle during pregnancy.(6)(7)(8)

The treatment for CMT can either be conservative or surgical, depending on how severe the condition is. If a diagnosis is delayed, it can lead to asymmetry in the head and cervical spine region. This asymmetry can cause imbalanced loads on the hip and pelvic joints, leading to deformities in other areas of the body. If left untreated, CMT can progressively limit head movement, resulting in eye movement disorders, craniofacial asymmetry, malocclusion, visual defects, neck pain, and a compensatory asymmetrical curvature of the spine that worsens with age.(9)

To sum up, the early detection of CMT in newborns and infants, coupled with timely treatment, provides an opportunity for complete resolution(10), and all the different possible complications' prevention.



I. <u>Patients:</u>

Our study is a retrospective conducted on 17 children with congenital torticollis. This is a series collected at the pediatric trauma-orthopedics department of the Mohammed VI University Hospital Center in Marrakech over a period of 14 years, starting from January 2009 to December 2023.

II. Inclusion criteria:

Included in our study :

- Patients with complete and usable medical records.
- Children with congenital torticollis operated at the pediatric trauma-orthopedics department of the Mohammed VI University Hospital Center in Marrakesh over a duration starting from January 2009 to December 2023.
- Age ≤ 15 years old.
- Follow up \geq 6 months.

III. Exclusion criteria:

Not included in this study were:

- Patients with incomplete and unusable medical records.
- Patients with non-congenital torticollis.
- Follow up < 6 months.

IV. Data collection :

For data collection, medical records of each patient were used to complete an exploitation form (appendix) aimed to achieve several objectives and purposes.

V. <u>Purposes of the study :</u>

- Epidemiological Understanding.
- Risk Factors Identification: Identify and analyze potential risk factors associated with the development of congenital torticollis in infants, contributing to a better understanding of its etiology.
- Diagnosis Accuracy: Assess the accuracy and reliability of diagnosis methods for congenital torticollis in infants, aiming to enhance early detection and intervention.
- Treatment Options and Effectiveness: Evaluate the effectiveness of various treatment modalities, including conservative approaches and surgical interventions, to determine optimal strategies for managing congenital torticollis.
- Functional and Quality of Life Outcomes: Examine the impact of congenital torticollis and its treatments on the functional abilities and overall quality of life of affected infants.
- Long-Term Follow-up: Conduct long-term follow-up assessments to understand the persistence, recurrence, or resolution of congenital torticollis as infants transition into childhood.
- Clinical Guidelines Development: Contribute to the development of evidence-based clinical guidelines for healthcare professionals involved in the care and management of infants with congenital torticollis.

By addressing these purposes, this study aims to enhance our knowledge of congenital torticollis, leading to improved diagnostic and therapeutic approaches. So ultimately, this research endeavors to delve into the nuances of surgical approaches for managing congenital

torticollis in infants which includes an in-depth exploration of surgical techniques, their effectiveness, potential complications, and overall outcomes. The goal is to provide a comprehensive understanding of the surgical and medical aspects in the management of congenital torticollis, aiding in the improvement of treatment strategies and patient outcomes.

VI. Statistical Analysis:

The statistical analysis of the data was performed using Microsoft Office Excel 2019 and google forms. Qualitative variables were expressed in percentages, and quantitative variables were expressed as means and ranges.

VII. <u>Ethics:</u>

Medical confidentiality and anonymity are respected in the exploitation forms.

VIII. <u>Surgical procedures :</u>

1. Unipolar tenotomy :

The release can be distal, releasing the SCM muscle attachments closer to the shoulder, or proximal, releasing the attachments closer to the skull.

The proximal unipolar tenotomy : the cephalic insertion should be meticulously dissected closest to the mastoid and precisely identified before the tenotomy; the main risk lies in the proximity to the facial and spinal nerves.

The unipolar low juxta-clavicular tenotomy technique involves accessing the lower insertions of the SCM muscle through a transverse incision positioned 1.5 to 2 cm above the clavicle.[Figure 1a]

Following dissection and protection of the external jugular vein [figure 2], the subcutaneous muscle is incised in the direction of the approach, and then the sheath of the SCM

muscle is opened. Both the clavicular [figure 3] and sternal [figure 4] heads are identified and sectioned using an electric scalpel. Due to the width of these lower attachments, multiple passes of the dissector are required to achieve complete tenotomy, allowing visualization of the deep plane (omohyoid muscle and internal jugular vein) [figure 5]. A 1 cm muscle resection helps to prevent recurrence. .[Figure 1b]

During surgery, passive mobilization of the head evaluates the outcome and addresses tension in residual fibers or a retracted aponeurosis, which should be liberally released. After ensuring hemostasis, reconstruction of the subcutaneous muscle plane is performed, followed by skin closure in two layers.

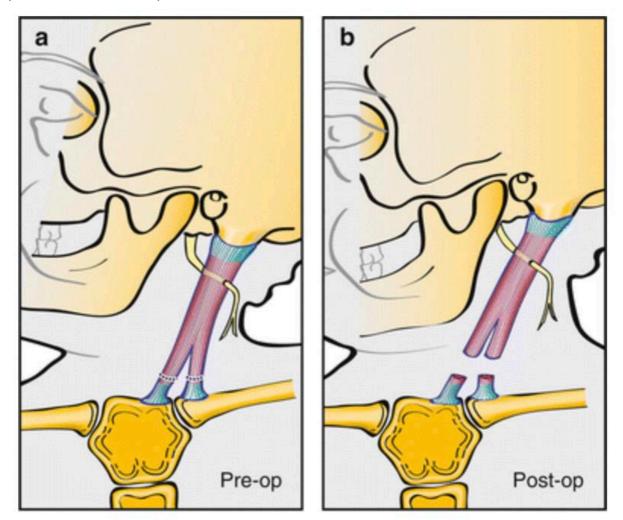


Figure 1: Distal unipolar tenotomy .a: pre-operative view. b; post-operative view.

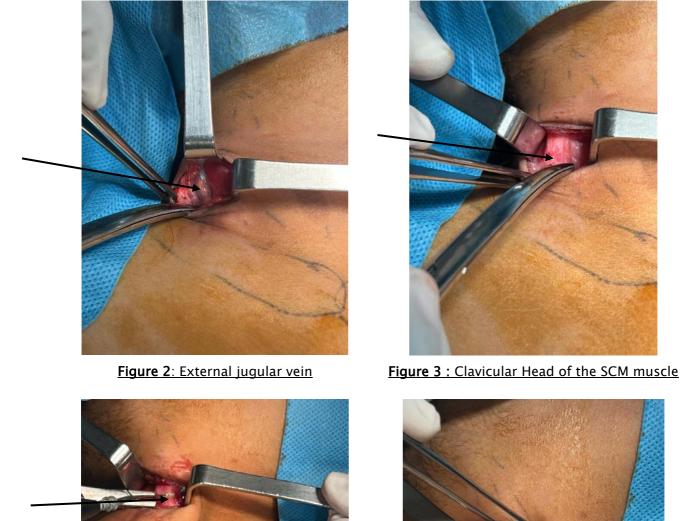




Figure 4: Sternal head of the SCM muscle.

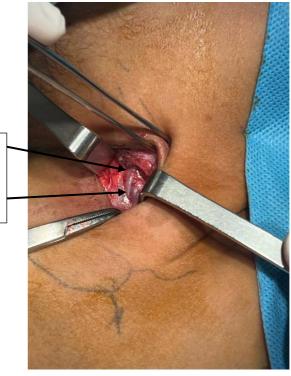


Figure 5: Deep plane (A :omohyoide muscle and B : internal jugular vein)

2. <u>Bipolar tenotomy :</u>

Described by Barcat. The technique involves making dual 1cm incisions, with sectioning of the proximal retro-auricular, mastoid insertion of the SCM muscle, and its distal supraclavicular insertion. The key concern is to locate and preserve the spinal nerve, which has a highly variable course.

The mastoid cranial portion is accessed through a short longitudinal incision centered on the muscular prominence behind the ear. Care must be taken not to extend this incision too far anteriorly into the cranial insertion to avoid injury to the facial and spinal nerves. After sectioning the subcutaneous neck muscle and cervical fascia, the muscular body is lifted onto a dissector and then sectioned with a cold scalpel under visual control. [Figure 6A]Complete correction of torticollis may require deep fascia sectioning, with caution to the auricular artery. [Figure 6B]

A variant of this cranial release involves a similar retro-auricular approach, where the mastoid periosteum is incised, and the entire SCM is detached subperiosteally, providing elongation without muscle dissection.

The caudal portion is approached with a horizontal incision just above the clavicle, centered on the insertion of the sternal and clavicular heads. [Figure 7A] The subcutaneous tissue and cervical fascia are sectioned, allowing dissection of the two muscle heads, which are then lifted and sectioned . If necessary, deep fascia sectioning reveals the proximity of vascular and nerve structures.[Figure 7B]

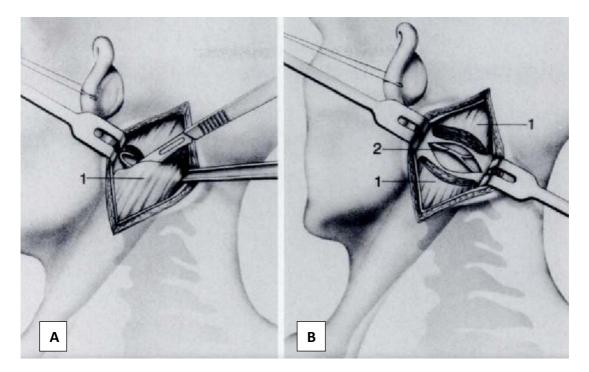


Figure 6: Sectioning of the proximal retro-auricular, mastoid insertion of the SCM. A: Mastoid insertion of the SCM, B; View after the SCM section.(1)SCM (2)auricular artery.

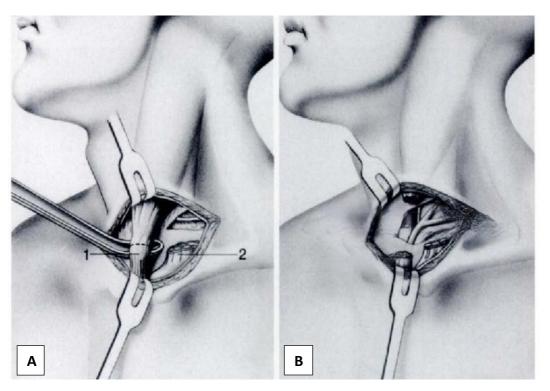


Figure 7: Sectioning of the SCM caudal portion. **A**: Sternal and clavicular SCM insertion. **B**:Deep fascia sectioning revealing vascular and nerve sturctures.(1) sternal insertion (2)clavicular insertion.

3. <u>Z-plasty:</u>

This technique, pioneered by Jones in 1923 and later popularized in France by Barcat and Dubousset, involves a horizontal incision placed within a neck fold at the midpoint of the muscle, where it is narrowest [figure 8]. After releasing all fascial attachments, the muscle is carefully sectioned with an electric scalpel in a stepwise manner from top to bottom and from inside to outside, ensuring protection of the spinal nerve.

It's crucial to cut the clavicular head as close to the clavicle as possible to anticipate the necessary muscle length for proper suturing post-correction [figure 9]. Typically, the upper clavicular head is sutured to its lower sternal counterpart to minimize tension [figure 10].

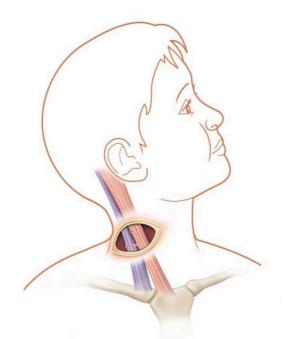


Figure 8: Z-plasty incision at the mid-third of the SCM where the heads are the closest and least thick.



Figure 9: The release involves both heads at different levels to achieve elongation and maintain muscular contour.

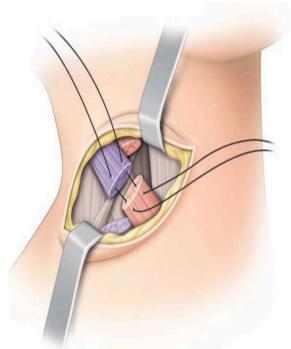


Figure 10 : the upper clavicular head is sutured to the lower sternal head to reconstruct a harmonious muscular contour.

IX. Evaluation Method:

The evaluation of results takes into account clinical, functional and cosmetic data. The criteria used are inspired by 2 scores:

| Excellent | No complaints, limitation of ROM of the neck, or facial deformity. | | | | | | |
|-----------|---|--|--|--|--|--|--|
| Good | Mild residual limitation of ROM of neck or mild residual facial deformity without complaints. | | | | | | |
| Fair | Residual limitation of ROM of neck and residual facial deformity without complaints. | | | | | | |
| Poor | Severe limitation of ROM of neck and obvious objective facial deformity, with complaints. | | | | | | |

Table I : Tanabe's assessment criteria (Grade Functional criteria)

Table II: Cheng and Tang's Scoring System for Assessment of Clinical and Subjective Outcome in Congenital Muscular Torticollis.

| Points | Rotational deficit | Lateral flexion deficit | Craniofacial asymmetry | Scar | Residual contracture | Subjective assessment (cosmetic and functional) | Head tilt |
|--------|-----------------------|-------------------------------|---------------------------|----------|-------------------------|---|------------------|
| 3 | $\leq 5^{\circ}$ | ≤ 5° | None | None | None | Excellent | $\leq 5^{\circ}$ |
| 2 | 6°-10° | 6°-10° | Mild | Mild | Lateral | Good | 6°-10° |
| 1 | 11°-15° | 11°-15° | Moderate | Moderate | Lateral, clavicular | Fair | 11°-15° |
| 0 | >15° | >15° | Severe | Severe | Clavicular, sternal | Poor | >15° |

• 17-21pts: Excellent 12-16pts: Good 7-11pts: Fair < 7 pts: Poor



I. Epidemiological data

1. Gender:

The distribution of patients in our study showed a slight male predominance, with 53.3% of patients being male and 46.7% being female. The sex ratio in our study, calculated as the ratio of males to females, stood at 1.14 indicating a slightly higher representation of males compared to females.

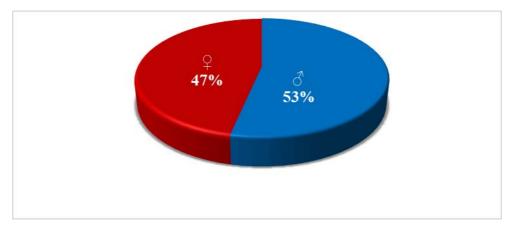
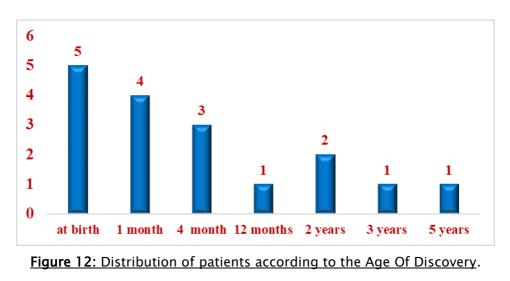


Figure 11: Distribution of patients according to Gender.

2. Age of discovery:

This distribution illustrated the ages at which the condition was discovered and officially diagnosed among the patients. The largest proportion of cases, 29.4%, were identified at birth, followed by 23.5% at one month, and 17.6% at four months. There was one case identified at 12 months, accounting for 5.9% of the total. Additionally, two cases (11.8%) were discovered at 2 years, and one case (5.9%) each at 3 years and 5 years, respectively.



3. Distribution by affected side:

This distribution illustrated a predominant occurrence of the condition on the right side, accounting for 70.6% of cases, while the left side represented 29.4% of cases.

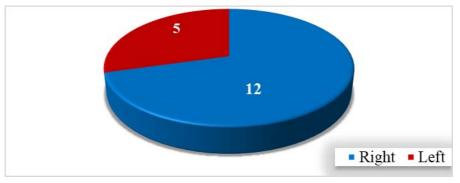


Figure 13: Distribution of patients according to the Affected Side.

II. Anamnesis :

1. History of the patients:

Eleven patients of 17 (64.7%) reported no particular history.

- 2 children were admitted in the case of a turner syndrome.
- 1 child from an inbreeding marriage survived prematurity and fetal distress

- 1 child presented a non- documented strabismus.
- 1 child noted a history of inguinal hernia surgery.

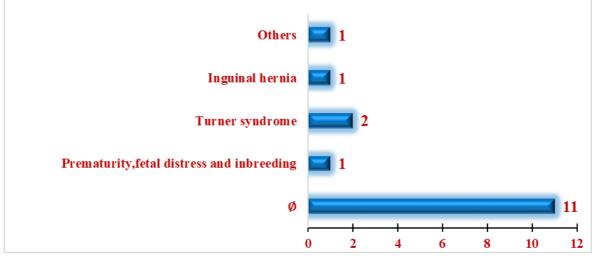


Figure 14: Distribution according to the History Of The Patient.

2. <u>Pregnancy and birth:</u>

2.1. Parity:

This distribution highlighted the parity status of mothers in the study population. 76.5% of cases were primigravida mothers , while 23.5% were multigravida mothers

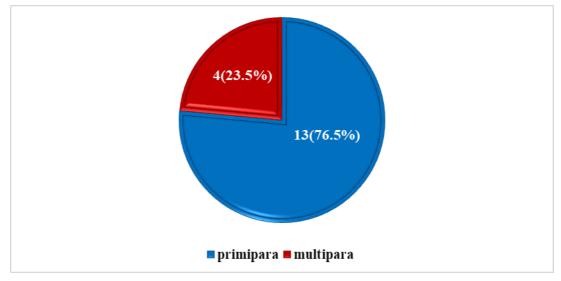


Figure 15: Distribution according to Parity.

2.2. Pregnancy follow-up:

This distribution underscored the variance in the level of prenatal care received by mothers in the study. 76.5% of cases, received a poor follow-up (few ultrasounds (less than one) and no regular quarterly consultations) during their pregnancies, potentially indicating inadequate access to or utilization of prenatal care services.

23.5% of mothers were classified as well-followed, suggesting that they received adequate prenatal care throughout their pregnancies.

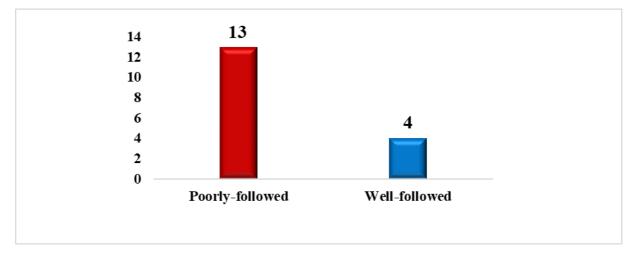


Figure 16 : Distribution according to the Pregnancy Follow-up.

2.3. <u>Multiple pregnancy:</u>

This distribution revealed the occurrence of multiple pregnancy within the study population. 11.8% of cases involved mothers who experienced a multiple pregnancy, indicating the presence of twins or higher-order multiples.

88.2% of cases, involved a singleton pregnancy.

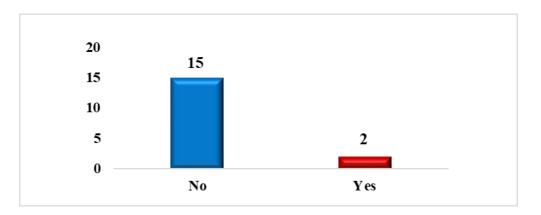
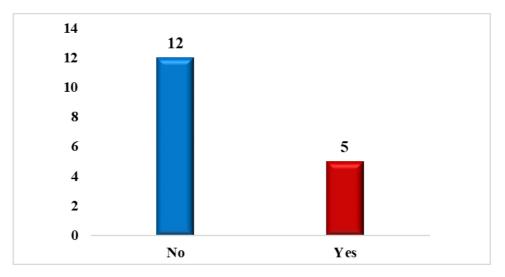
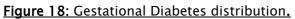


Figure 17: Multiple Pregnancy distribution.

2.4. Gestational diabetes:

This distribution highlighted the prevalence of gestational diabetes within the study population. 29.4% of cases involved mothers who experienced gestational diabetes during pregnancy, while 70.6% of cases, did not.





2.5. Term:

94.1% of all the cases noted full term pregnancies and only 5.9% prematurity which translated as only 1 child amongst 17.

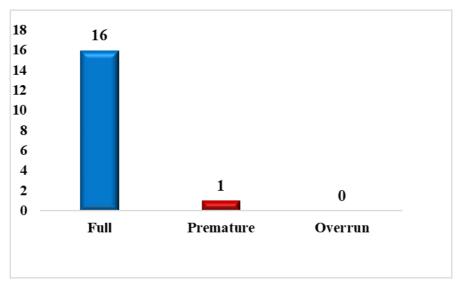


Figure 19: Pregnancy's Term distribution.

2.6. <u>Labor:</u>

This distribution offered insight into the labor experiences of the children in our study. 59% of cases, underwent labor without significant complications, following a typical progression of childbirth.

41% of cases faced challenges during labor, characterized by complications and prolonged duration.

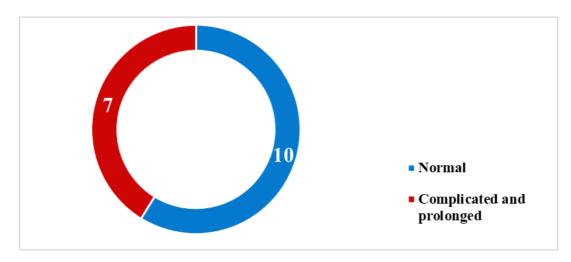


Figure 20 : Pregnancy Labor Progress distribution.

2.7. <u>Delivery presentation:</u>

This distribution provided insight into the fetal positioning within the study cohort. 52.9% of cases, presented in the breech position.

47.1% of cases were cephalic, indicating the head-first presentation, which is considered the optimal position for childbirth. Notably, there were no cases of transverse positioning.

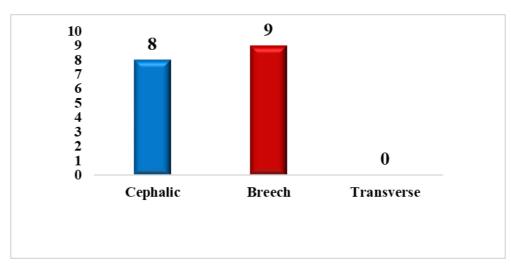


Figure 21: Delivery Presentation distribution.

2.8. Delivery:

This distribution revealed the diversity of delivery methods employed within our study. 29.4% of deliveries were conducted through natural birth, representing spontaneous vaginal delivery without the need for medical interventions. Similarly, another 29.4% of deliveries were carried out via Caesarean section, which may have been indicated due to various maternal or fetal factors necessitating surgical delivery. Additionally, 35.3% of deliveries required the use of instruments, such as forceps or vacuum extraction, to assist with the delivery process. Finally, 5.9% of deliveries involved an episiotomy.

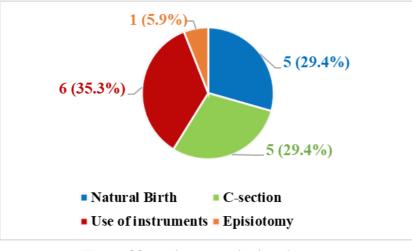


Figure 22 : Delivery Mode distribution.

2.9. Birthweight:

Unfortunately we couldn't collect the data about the birthweight of the patients of our study.

III. <u>Clinical examination:</u>

1. Head tilt and head rotation limitation:

All of the patients presented a head tilt of the affected side alongside with a limited head rotation of which 59% came with a $>15^{\circ}$ limitation and 41% with a $\leq 15^{\circ}$ limitation.

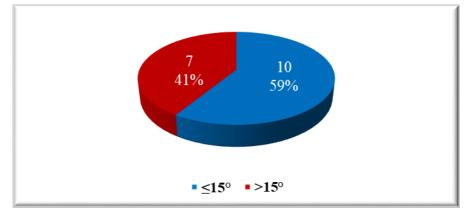
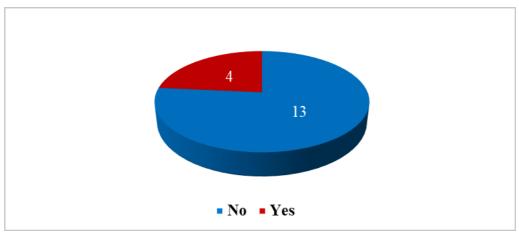


Figure 23: Head Rotation Limitation Degree distribution.

2. Facial asymmetry and plagiocephaly :

This distribution provided insight into the prevalence of facial asymmetry among the study cohort with congenital torticollis. 76.5% of cases, did not present a facial asymmetry. However, 23.5% of cases exhibited facial asymmetry.



All of the patients exhibited no plagiocephaly.

Figure 24: Facial Asymmetry distribution.

3. Shoulder elevation of the opposite side :

This distribution shed light on the prevalence of shoulder elevation on the contralateral side among the children with congenital torticollis in our study.

52.9% of cases, did not present a shoulder elevation on the contralateral side. However, 47.1% of cases exhibited one.

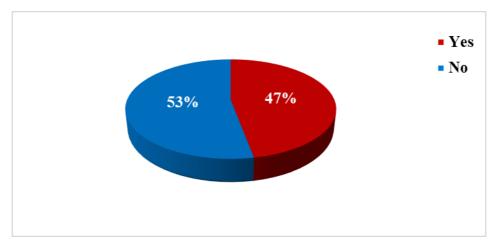


Figure 25: Shoulder Elevation Of The Contralateral Side distribution.

4. Olive/SCM Fibrotic mass :

Fourteen (82.4%) cases showed no evidence of an olive/SCM fibrotic mass, while 3 (17.6%) cases exhibited such a mass.

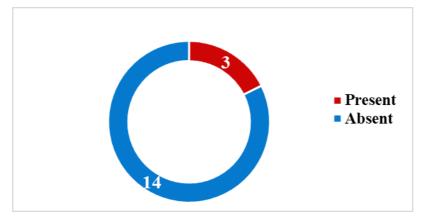


Figure 26: Presence of an Olive/SCM Fibrotic Mass.

5. <u>Hip examination :</u>

All of our patients presented a normal clinical hip examination.

6. Foot deformities:

88% of cases showed no association between the presence of a foot deformity in association with congenital torticollis , while 12% presented with one especially a flat foot.

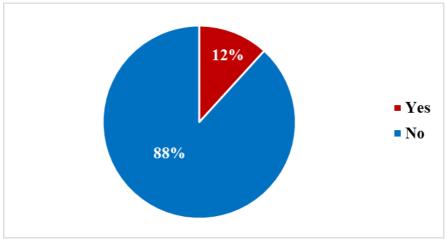


Figure 27: Foot Deformities distribution.

7. Scoliosis:

64.7% of cases showed no association between scoliosis and congenital torticollis, while 35.3% exhibited a dorso-lumbar scoliosis.

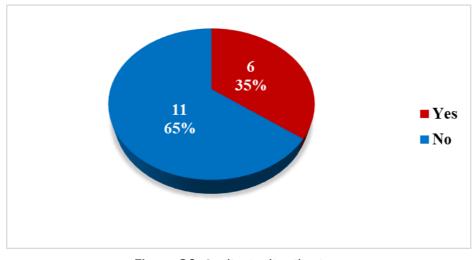


Figure 28: Scoliosis distribution.

IV. Paraclinical examination:

1. Ultrasound:

Amongst 17 patients only one benefited of an ultrasound examination showing a fibrotic olive.

2. Standard X-ray:

Nine (52.9%) of the 17 children underwent Standard X-ray that showed a deviation of the cervical spine to the affected side while 47.1% of all the cases have not benefited of one.



Figure 29: Standard X-ray of the front cervical spine performed as part of the initial work-up for congenital torticollis in a 6 years old child.

3. <u>CT-scan:</u>

Thirteen patients didn't undergo a CT-scan nor, while only 2 benefited from a CT-scan which showed a scoliosis with a convexity of the affected side and straightening of the cervical spine.

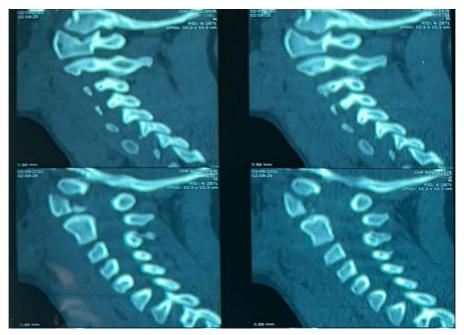


Figure 30 : Sagittal CT-scan section of the cervical spine performed to rule out a vertebral anomaly in a 7 years old child with congenital torticollis.

4. <u>MRI:</u>

Only one of the patients underwent an MRI examination which showed a D12-L1 block vertebra.



Figure 31: MRI of the cervical spine showing a normal spinal canal with no vertebral anomalies of a 6 years old child.

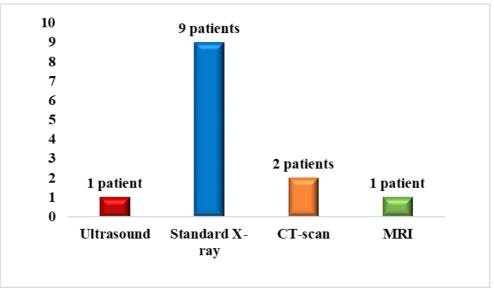


Figure 32: The Paraclinical Tests prescribed for the patients.

V. <u>Treatment:</u>

1. <u>Pre-operative physiotherapy:</u>

1.1. Duration:

- 29.4% received treatment for 2 months
- 47.1% for 3 months
- 17.6% for 6 months
- 5.9% for 1 year

But all of these duration has showed no significant efficiency.

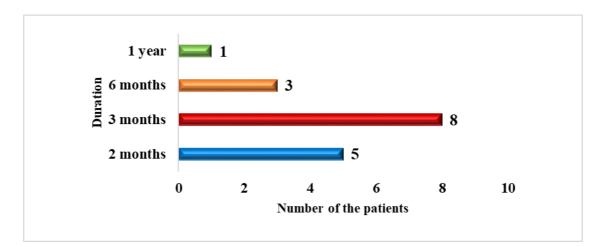


Figure 33: Pre-operative Physiotherapy duration.

1.2. Frequency:

- Three went for a once a week program
- Twelve children adopted a 2 times a week physiotherapy programs.
- Two opted for a 3 times a week regimen.

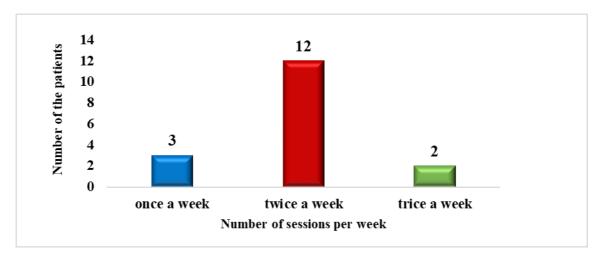


Figure 34: Pre-operative Physiotherapy Frequency.

1.3. <u>Pre-operative physiotherapy age debut:</u>

6 of the patients started the physiotherapy at 3 years old, 2 at 3 months old ,another 2 at 2 years old, and another 2 at 9 years old, 1 child respectively at 4 months old, 8 months old , 9 months old , 6 years old and 8 years old.

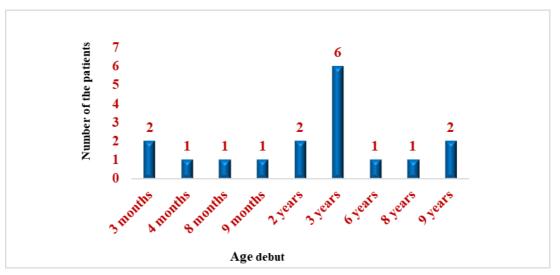


Figure 35: Pre-operative Physiotherapy Age Debut.

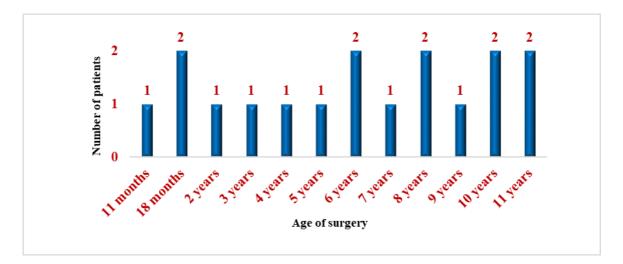
2. Surgery:

2.1. Age of surgery:

The distribution of ages at the time of surgery revealed variability in the timing of surgical intervention for congenital torticollis. The most common ages for surgery were 10 years and 11 years, each representing 11.8% of cases. This suggests that surgical correction is often performed during late childhood or early adolescence.

Additionally, surgeries were also performed at younger ages, with 11 months and 18 months each representing 5.9% of cases. This indicates that some patients underwent surgical intervention during infancy or toddlerhood.

We can come to the conclusion that the 2/3 of our patients underwent surgery at an age ≥ 6 years old.



2.2. Duration:

The average duration of surgeries is 39.71 minutes with extremes going from 20 minutes to 65 minutes.

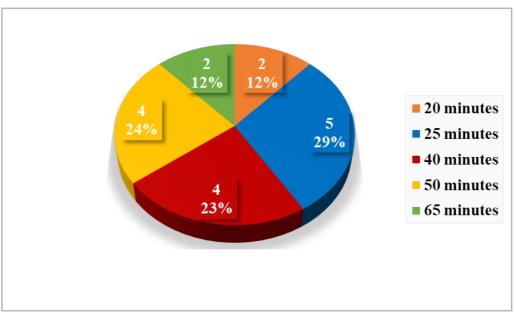


Figure 37: Surgery Duration.

2.3. Position:

All of the patients were in dorsal decubitus during their surgery.

2.4. Technique used:

82.4% of the patients underwent a unipolar tenotomy, 11.8% a bipolar tenotomy and 5.9% a Z-plasty.

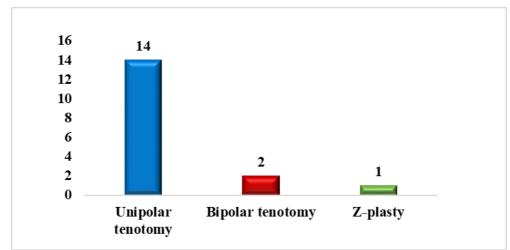
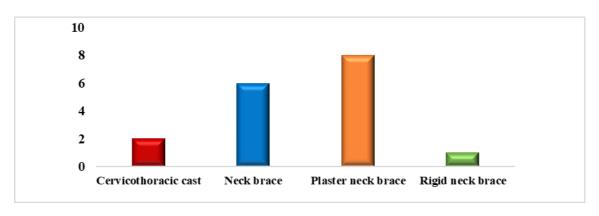


Figure 38: Type The Surgery Procedure.

3. Post-operative management:

3.1. Means of immobilization:

47,1% of the patients were prescribed a plaster neck brace, 35,3% a regular neck brace, 11.8% a cervicothoracic cast and 5.9% a rigid neck brace.





3.2. Duration of immobilization:

The average duration of post-operative immobilization was 45.94 days approximately with extremes going from 4 days to 6 months with a discontinuous wear of 12 hours per day.

P.S: the 4 days post-operative immobilization is strictly due to a non-compliance of the duration prescribed because of the child's discomfort and refusal of wearing the orthosis.

The 2 patients that wore the orthosis for 6 months, wore them continuously for a duration of 4 weeks then switched to a discontinuous wear of an 8 to12 hours per day nightly time.

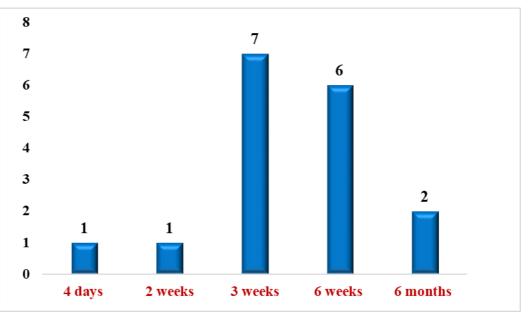


Figure 40 : Immobilization Duration.

3.3. <u>Resumption date of physiotherapy:</u>

The average resumption date of post-operative physiotherapy was 12.24 days with extremes extending from a week to a month post-surgery, and a redisplay of the immobilization mean after physiotherapy.

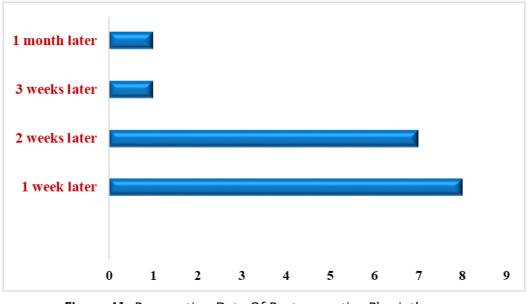


Figure 41: Resumption Date Of Post-operative Physiotherapy.

3.4. <u>Duration of post-operative physiotherapy:</u>

Seven patients underwent 3 months of post-operative physiotherapy, 6 encountered 6 months and 4 children only did 2 months of post-surgery physiotherapy. That resulted in an average of 3.82 months.

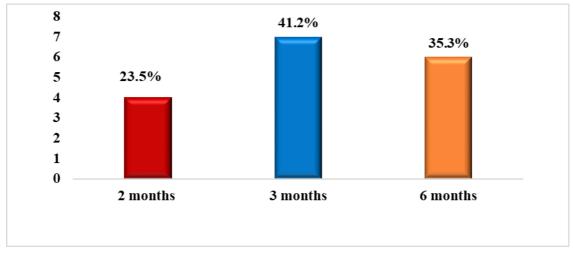


Figure 42: Post-operative Physiotherapy Duration.

3.5. Frequency of post-operative physiotherapy:

76.5% of the study cases went through a twice a week post-operative schedule while the 23.5% left experienced a trice a week program.

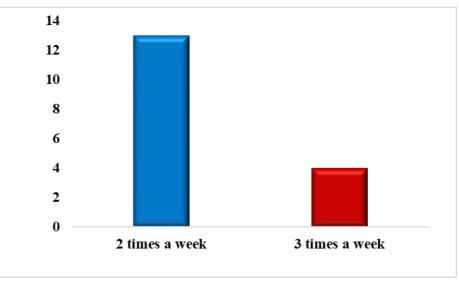


Figure 43: Post-operative Physiotherapy Frequency.

VI. <u>Results:</u>

1. Tanabe's assessment criteria:

88.2% of the patients demonstrated excellent outcomes according to Tanabe's assessment criteria. This indicated a high level of success or effectiveness in the treatment or intervention provided. Additionally, 11.8% of cases showed good outcomes, suggesting positive but slightly less optimal results compared to the excellent category. Overall, the distribution of outcomes reflects a predominantly favorable response to the intervention based on Tanabe's assessment criteria, highlighting its efficacy in managing the condition under study.

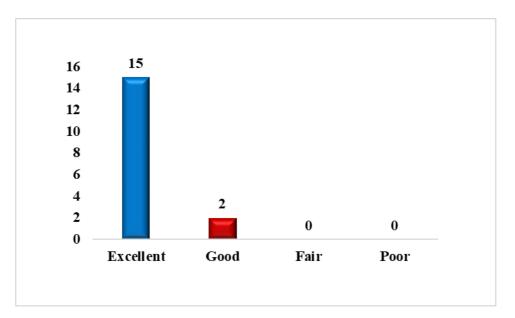


Figure 44: Results according to Tanabe's Assessment Criteria.

2. Cheng and Tang's score:

The distribution of outcomes based on the Cheng and Tang's Score indicated predominantly positive outcomes within the studied group. Specifically, 70.6% of cases achieved an excellent rating, indicating highly successful outcomes based on the scoring system. Additionally, 29.4% of cases received a good rating, suggesting favorable outcomes though not reaching the highest level observed. Notably, no cases were rated as fair or poor, implying a lack of significant limitations in the outcomes assessed. Overall, the findings highlight the effectiveness of interventions or treatments evaluated using this scoring system, with the majority demonstrating excellent or good outcomes.

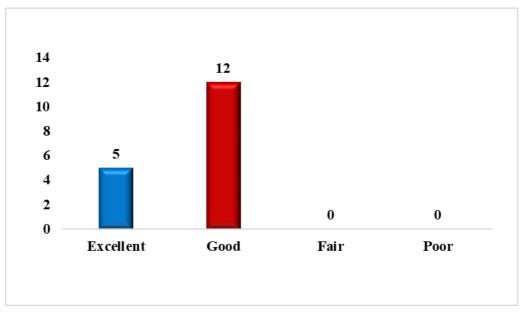


Figure 45: Results according to Cheng and Tang's Score.

3. <u>Results according to age:</u>

- 1.1. Tanabe's assessment criteria:
 - <u>Excellent Outcome:</u> 15 of the surgeries resulted in an excellent outcome according to Tanabe's assessment criteria. Patients who underwent surgery at ages ranging from 11 months to 11 years achieved excellent outcomes.
 - <u>Good Outcome</u>: There were two cases where the surgery resulted in a good outcome. Both of these cases were operated at 6 years and 5 years of age, respectively.

Overall, the majority of surgeries across different age groups resulted in excellent outcomes, indicating the effectiveness of the surgical interventions in treating the condition.

| Age of Surgery | Excellent | Good |
|-----------------|-----------|------|
| 11 months (n=1) | 1 | 0 |
| 18 months (n=2) | 2 | 0 |
| 3 years (n=2) | 2 | 0 |
| 4 years (n=1) | 1 | 0 |
| 5 years (n=2) | 1 | 1 |
| 6 years (n=1) | 0 | 1 |
| 7 years (n=1) | 1 | 0 |
| 8 years (n=2) | 2 | 0 |
| 9 years (n=1) | 1 | 0 |
| 10 years (n=2) | 2 | 0 |
| 11 years (n=1) | 1 | 0 |
| Total (n=17) | 15 | 2 |

Table III:Distribution of Tanabe's assessment criteria outcomes according to the age of surgery.

1.2. Cheng and Tang's score:

- Excellent Outcome: 5 of the surgeries resulted in an excellent outcome according to Cheng and Tang, these surgeries were performed at 11 months, 18 months, 3 years (only one of the 2 surgeries performed at this age) and 4 years.
- <u>Good Outcome</u>: a total of 12 surgeries , spanning ages from 3 years to 11 years, resulted in good outcomes of age achieved good outcomes based on Cheng and Tang scoring system.

Overall, surgeries across various age groups resulted in either excellent or good outcomes according to the Cheng and Tang, indicating the effectiveness of the surgical interventions.

| Age of Surgery | Excellent | Good |
|-----------------|-----------|------|
| 11 months (n=1) | 1 | 0 |
| 18 months (n=2) | 2 | 0 |
| 3 years (n=2) | 1 | 1 |
| 4 years (n=1) | 1 | 0 |
| 5 years (n=2) | 0 | 2 |
| 6 years (n=1) | 0 | 1 |
| 7 years (n=1) | 0 | 1 |
| 8 years (n=2) | 0 | 2 |
| 9 years (n=1) | 0 | 1 |
| 10 years (n=2) | 0 | 2 |
| 11 years (n=2) | 0 | 2 |
| Total (n=17) | 5 | 12 |

Table IV: Distribution of Cheng and Tang's Score outcomes according to the age of surgery

4. Results according to the technique used :

4.1. Tanabe's assessment criteria:

- <u>Unipolar Technique</u>: Out of the 13 cases treated with the unipolar technique, 11 (100%) achieved an excellent outcome according to Tanabe's assessment criteria.
- <u>Bipolar Technique:</u> For the 3 cases treated with the bipolar technique, 2 (66.7%) achieved a good outcome, while 1 (33.3%) achieved an excellent outcome according to Tanabe's assessment criteria.
- <u>Z-plasty Technique</u>: The single case treated with the Z-plasty technique achieved an excellent outcome according to Tanabe's assessment criteria. However, due to the limited data, it's challenging to draw definitive conclusions about the effectiveness of this technique compared to others.

Overall, all the cases across all techniques achieved excellent or good outcomes according to Tanabe's assessment criteria, suggesting overall positive results in the treatment of the condition.

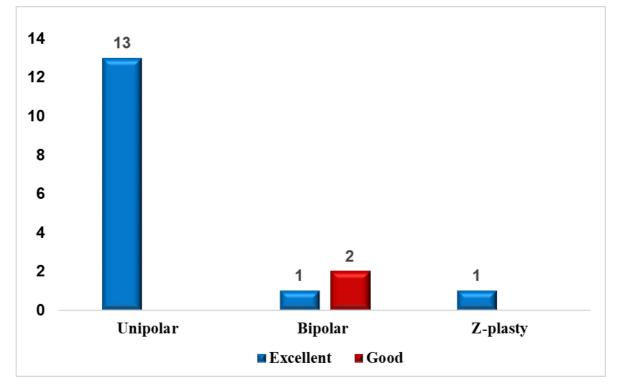


Figure 46: Distribution of Tanabe's assessment criteria outcomes according to the technique used.

4.2. Chang and Tang score:

- <u>Unipolar Technique</u>: Out of the 13 cases treated with the unipolar technique, 10 (72.7%) achieved a good outcome, while 3 (27.3%) achieved an excellent outcome according to Cheng and Tang's score.
- <u>Bipolar Technique:</u> For the 3 cases treated with the bipolar technique, all 3 (100%) achieved a good outcome according to Cheng and Tang's score.
- <u>Z-plasty Technique</u>: The single case treated with the Z-plasty technique achieved an excellent outcome according to Cheng and Tang's score. However, due to the limited data, it's challenging to draw definitive conclusions about the effectiveness of this technique compared to others.

Overall, all of cases across all techniques achieved a good or excellent outcome according to Cheng and Tang's score, suggesting overall positive results in the treatment of the condition.



Figure 47 : Distribution of Cheng and Tang's Score outcomes according to the technique used.



Figure 48: Unipolar tenotomy results in a 5 year-old patient.



Figure 49: Bipolar tenotomy results in a 6 year-old patient.



Figure 50:The evolution of a girl case in our department with excellent results of a 1 month follow-up after a unipolar tenotomy.



Figure 51: Excellent results of the same patient after a 6 months follow-up.

VII. <u>Follow-up:</u>

The average follow-up was 7.35 months with extremes going from 6 months to 15 years.



I. <u>History overview:</u>

Taylor (1875) first described the pathology of CMT from an autopsy specimen of a 6week-old baby as "an induration of the sterno-mastoid muscle or sterno-mastoid tumour,"(11)

The historical roots of congenital torticollis trace back to ancient times. Antyllus, known for performing tenotomy for torticollis in the second century, as documented by Oribasius a century later (Hulbert 1950). MacDonald (1969) categorized CMT into sternomastoid tumor and muscular torticollis groups based on the presence of SCM tightness or tumor. Hulbert (1950) introduced the term postural torticollis for cases without SCM tightness or tumor. However, the distinction between postural torticollis and CMT has not been consistently defined in literature, with CMT often used interchangeably with congenital torticollis in most studies. (11)

II. Anatomical overview:

Congenital torticollis -wryneck- is a condition resulting from the cervical muscles retraction. The SCM muscle is the main culprit, but other muscles such as the scalenes, the trapezius upper portion, the splenius of the head and a whole series of paravertebral muscles can also contribute to its occurrence. A thorough knowledge of the anatomy of the SCM, including the distribution and arrangement of its various muscle bundles, is essential to understanding the complexity of head movements, including lateral tilt, rotation and translation, as well as the pathophysiology of torticollis.

1. The SCM muscle:

The SCM muscle is one amongst more than 20 pairs of muscles influencing the neck. It receives innervation from two different nerves and serves various functions. It is easily palpable on the surface, hence the significance it holds as a key anatomical reference point in the neck area

and the important role it plays in conditions involving neuromuscular issues like torticollis. (12)

The muscle begins its course from the upper edge of the sternal manubrium and the medial quarter of the upper aspect of the clavicle. These two muscle heads converge to form a unified muscle belly that ascends upwards and laterally. Its attachments extend to the mastoid process of the temporal bone and the front section of the superior nuchal line. The SCM features fibers aligned in parallel fashion; it does not possess a pennate muscle structure.(13)

1.1. Superficial plane :

Composed of 3 heads:

- <u>The cleido-occipital head</u>:
 - ✓ ORIGIN:

It attaches to the upper third of the superior surface of the clavicle, near its posterior border, via short tendinous fibers. It is positioned behind and parallel to the posterior border of the sterno-occipital head.

✓ PATH:

It ascends upwards, backwards, and outwards, presenting a rectangular shape that largely covers the cleidoic-mastoid.

✓ TERMINATION:

It ends in an aponeurotic blade on the lateral two-thirds of the superior nuchal line, inside the sterno-occipital head.

• <u>The sterno-occipital head:</u>

✓ ORIGIN:

It attaches with a short tendon, combined with that of the sterno-mastoid head, on the anterior surface of the sternal manubrium, near the articular facet of the clavicle.

✓ PATH:

It ascends upwards, backwards, and outwards, appearing as a very thin structure, peripheral to the posterior border of the sterno-mastoid, located externally to it.

✓ TERMINATION:

It ends in a small tendinous blade on the lateral part of the superior nuchal line, outside the cleidoic-occipital head.

- <u>The sterno-mastoid head:</u>
 - ✓ ORIGIN:

It shares a common origin with the sterno-occipital head, inserting with a robust tendon on the anterior surface of the sternal manubrium, below the sternal notch and below and inside the sternocostoclavicular joint.

✓ PATH:

It ascends upwards, backwards, and outwards, presenting an elongated, triangular shape with a lower apex. Its lower part is cylindrical for two-thirds, then becomes kidney-shaped and flattened for the upper third.

✓ TERMINATION:

It ends in an aponeurotic blade on the lateral aspect of the mastoid process.

1.2. Deep plane :

It consists of a unique cleido-mastoid head.

✓ ORIGIN:

Located behind the cleido-occipital head.

✓ PATH:

Running vertically or slightly obliquely upwards and backwards. As a result, it is initially concealed by the cleidoic-occipital head, then by the sterno-mastoid head of the superficial plane.

✓ TERMINATION:

At the mastoid process, along a line parallel and inferior to that of the sterno-mastoid head.

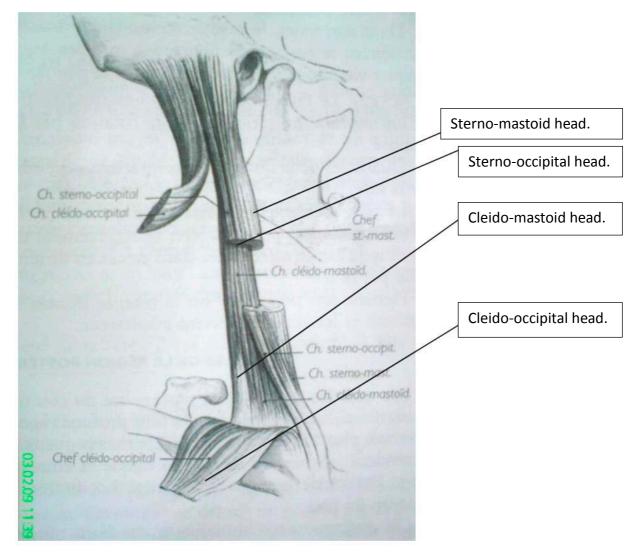


Figure 52: SCM heads.

1.3. Functions :

The unilateral contraction of the SCM results in three movements: rotation of the head towards the opposite side of its contraction, inclination towards the side of its contraction, and extension.(12)

The SCM can also act as an inspiratory muscle by anchoring to a fixed point on the temporal bone and subsequently raising the sternum and clavicles.(12)

The SCM is pivotal in maintaining both neck and body posture. Studies have demonstrated that vestibular stimulation can electrically trigger the SCM, highlighting a strong link between the vestibular region and the motoneurons governing the SCM.(14)

Lateral inclination is the motion where the SCM demonstrates its highest capability for speed and force.(15)

Another crucial role of the SCM is facilitating proper function of the temporomandibular joint.

1.4. Vascularization:

a. Vessels :

The blood supply of the SCM is derived from 3 principal arteries :

- The superior thyroid artery that originates from the external carotid artery.
- The superior scapular artery that arises from the sub clavicular artery branch.
- The sternocleidomastoid artery which is a branch of the occipital artery.

The external jugular vein courses downwards and backwards behind the SCM muscle, collecting venous blood from the external posterior and anterior jugular veins.(12)

b. Lymphatics:

The lymphatic system responsible for draining the SCM muscle is known as the vertical chain, comprising the anterior superficial lymph nodes and the lymph nodes located in the inferior part of the posterior triangle.(12)

1.5. E.Innervation

The cervical plexus cutaneous branches emerge from the posterior border of the SCM muscle, providing nerve endings that aid in its proprioceptive functions. Additionally, the

accessory nerve (cranial nerve XI) travels through the posterior triangle to supply innervation to both the trapezius and the sternocleidomastoid.

1.6. General anatomical relationships:

The cervical spine:

The skeletal structure of the neck comprises 7 bony elements: the cervical vertebrae. The hyoid bone is often included due to its close association with the soft tissues of the anterior neck region.

The concept of anatomical and functional division of the cervical spine has led to the distinction of 2 opposing levels:

- The upper level, consisting of the atlas and axis, is responsible for rotational movements.
- The lower level (from C3 to C7) is dedicated to flexion and extension. The greatest mobility of the cervical spine is found in the lower segment.

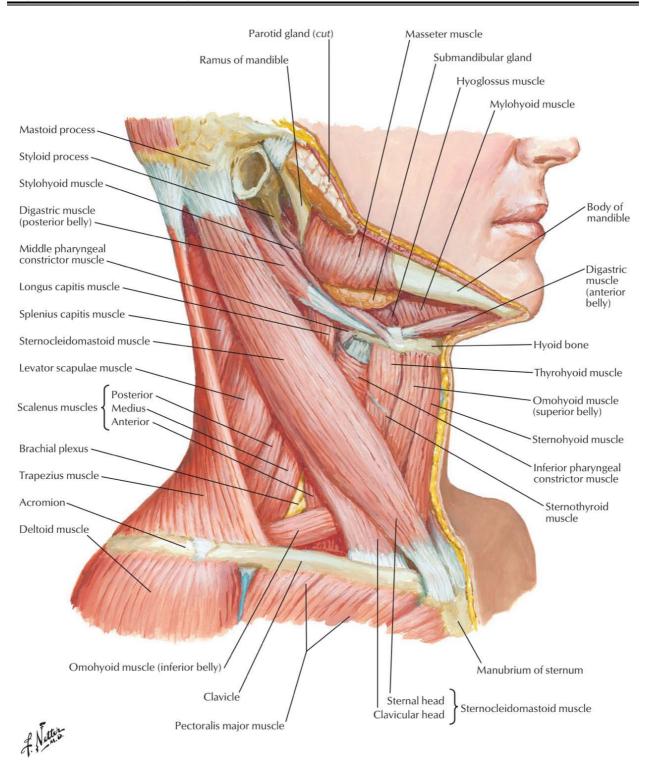


Figure 53: Anatomy of the Neck. (Netter).

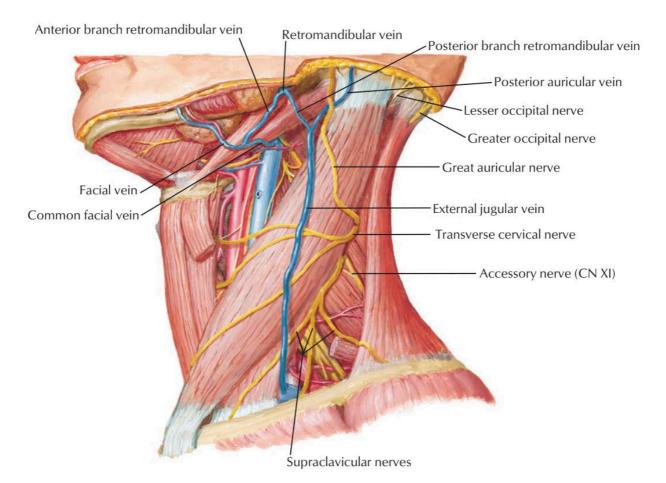


Figure 54: Lateral view of the muscles of the neck.

III. Embryology overview :

The SCM muscle originates from both paraxial (pre-optic) mesoderm and occipital (post-optic) somites, with some contribution from the neural crests(16) (17). Its development initiates around the 14th day of gestation in animal models. Recent research suggests that the precursor cells of neck muscles coexist with cardiac progenitor cells in the cardiopharyngeal mesoderm.(18)

IV. Physiologic variants:

Congenital absence of the SCM muscle, which might sometimes involve the simultaneous absence of the trapezius muscle, is a rare anomaly that may not result in noticeable clinical or functional impairments. This is likely because other muscles in the neck region compensate for the missing muscles through adaptive changes.(19)

Different variations of the SCM muscle can influence surgical procedures in the area, particularly its origin. The attachment to the clavicle may vary in width, ranging from narrow to wide (up to approximately 7 to 8 cm), or there may be multiple clavicular attachments. These variations could also impact the acromioclavicular joint or result in additional muscular bellies within the SCM muscle. Some variations involve insertions into the sternoclavicular joint, altering the neck's anatomy.(20)

An increased number of SCM muscle heads is relatively uncommon. For instance, one side may exhibit two sternomastoid heads, along with a cleido-occipital and a cleidomastoid occipital origin, while the other side may have a single sternomastoid head, a cleido-occipital origin, and two cleidomastoid origins, resulting in a total of four muscle heads.(21)

Occasionally, the border of the SCM muscle may directly interface with the trapezius, likely stemming from embryological abnormalities. Variants such as cleido-epistrophic, cleidocervical, and cleido-atlantic insertions are recognized, each characterized by one or more heads.

The innervation of the SCM muscle can exhibit variability. According to a study, the lower part of the SCM may receive innervation from a branch of C1 originating from the ansa cervicalis (descendens hypoglossi); likewise, this pattern may occur exclusively for the upper part of the muscle(22). Additionally, an aberrant branch of the facial nerve has been identified to innervate the deep portion of the upper third of the SCM.(23)

Given the range of anatomical variations outlined, it is advisable to maintain high caution when approaching the area for surgical procedures.

V. Anatomopathology overview:

The histological structure of the (SCM) muscle in individuals with congenital torticollis exhibits distinct changes due to the condition. Typically, congenital torticollis is associated with unilateral shortening and thickening of the SCM muscle, accompanied by fibrosis and collagen deposition. This results in a noticeable and palpable mass in the affected muscle, causing the characteristic head tilt and rotation seen in these individuals. Histopathological examination often reveals endomysial fibrosis, a diffuse proliferation of uniform fibroblasts and myofibroblasts, degenerative skeletal muscle fibers, and positive staining for Vimentin and actins within the affected SCM muscle. These changes contribute to the restricted range of motion, muscle imbalance, and structural abnormalities associated with congenital torticollis. Consequently, the histological structure of the SCM muscle in individuals with congenital torticollis is marked by fibrosis, collagen deposition, and specific cellular alterations that differ from those in individuals without the condition.(24) [figure 51]

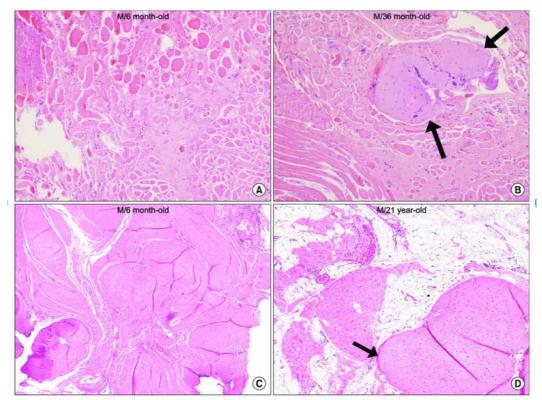


Figure 55: Histopathological findings of the SCM.

The histopathological findings of the sternocleidomastoid muscle with congenital muscular torticollis. (A) Diffuse interstitial fibrosis with accompanying atrophic muscle fibers is noted (H&E, \times 200). (B) Interstitial fibrosis with presence of aberrant tendon-like excessive dense connective tissue (arrows) (H&E, \times 200). (C) Interstitial fibrosis with presence of aberrant tendon-like excessive dense connective tissue which was well-arranged (H&E, \times 40). (D) Aberrant tendon-like excessive dense connective tissue and prominent fat infiltration (arrow) (H&E, \times 40).(25)

Macroscopic Aspect :

- Unilateral shortening and thickening of the affected SCM muscle.(25)(26)
- Presence of a palpable and visible mass in the SCM muscle. (25)
- The SCM muscle appears thickened and condensed along its length, leading to limited neck rotation and lateral flexion.(24)

Microscopic Aspect :

- Endomysial fibrosis with collagen deposition and accumulation of uniform fibroblasts and myofibroblasts.(24)
- Degeneration of skeletal muscle fibers. (24)
- Positive staining for vimentin and actins. (24)
- Diffuse proliferation of uniform fibroblasts and myofibroblasts. (25)
- No macroscopic hemorrhage or necrosis.(24)

These characteristic histological changes contribute to the limited mobility, muscle imbalance, and structural abnormalities observed in congenital torticollis.(25) They differ from the normal histology of the SCM muscle.

VI. <u>Physiopathology:</u>

The physiopathology of (CMT) involves structural changes in the (SCM) muscle, leading to unilateral shortening and fibrosis.

The exact etiology remains unclear, but several theories exist:

- Birth trauma.(27)
- Prenatal or perinatal compartment syndrome.(28)
- Impairment of the developing sternocleidomastoid (SCM) muscle due to intrauterine constraint.(7)(29)
- → Recent studies, particularly immunohistochemical and gene expression analyses, provide stronger evidence for an intrauterine impairment of the developing SCM as the primary cause of CMT.(7)(8)
- → The main pathological features of the affected SCM muscle include:(8)
 - Excessive endomysial and perimysial fibrosis.
 - Adipocyte hyperplasia.
 - Muscle fiber atrophy.
- → These changes lead to tightness of the involved SCM and limited cervical motion. The degree of fibrosis is directly proportional to the age of the child when left untreated; older children typically exhibit more fibrosis.(8)
- → Emerging immunohistochemical research suggests that the deposition of type III collagen is a key factor in generating SCM fibrosis, and its hyperplasia is associated with accelerated apoptosis and overexpression of transforming growth factor beta 1.(30)
- → Gene expression studies support the notion that fibrosis with collagen and elastin fibrillogenesis is one of the critical pathways in the pathogenesis of CMT. These studies also provide evidence of DNA repair and cytoskeletal rearrangement, possibly related to mechanical strain.(31)

In summary, while the exact etiology of CMT remains unclear, recent research points towards an intrauterine impairment of the developing SCM muscle as the primary cause, leading to excessive fibrosis, adipocyte hyperplasia, and muscle fiber atrophy, which ultimately result in the characteristic features of CMT.

VII. Epidemiological data :

1. <u>Gender :</u>

According to both studies Cheng(11)(32)and Petronic (33), a male predominance exists with a sex-ratio of 3/2. In our series we have noted a slight male predominance with a ration of 1.14.

2. <u>Age :</u>

Understanding the age at which the condition is typically discovered is an essential determinant of the timing of diagnosis and potential early interventions.

The mean age of CMT in different studies goes from 2 to 6 months, while our study has shown the mean age was 10.12 months with extremes going from an at birth discovery to 5 years.

| Series | Country | Mean age |
|--|-----------|--------------|
| Ho et al.(27) | Singapore | 2 months |
| Bastos et al. (34) | Portugal | 6 months |
| Amaral et al.(7)7/26/2024 10:42:00 AM | Portugal | 3 months |
| Cheng et al. (40) | Hong Kong | 2.3 months |
| AbdurRahman LO et al.(36) | Nigeria | 3 months |
| Our study | Morocco | 10.12 months |

Table V: Comparison of average ages by study

3. Affected side :

Multiple studies (Cheng et al(6)., Petronic et al(5)., Xu et al.(33))consistently show that CMT has a higher prevalence of affecting the right side of the neck compared to the left side, with the right side being involved in approximately 60–61% of cases which aligns with the results of our study. The bilateral affection exists but remains rare, with an incidence estimated at only 0.3 to 2% of congenital torticollis cases. (34) (37)

VIII. <u>Risk factors :</u>

The key risk factors for congenital muscular torticollis (CMT) include:

- Male gender.
- Primiparity.
- Birth trauma:
 - Large birth weight or macrosomia.
 - Breech presentation.
 - Multiple pregnancy.
 - Difficult and long labor and delivery.
 - Use of instrument such as vacuum or forceps.
- Intrauterine Factors:
 - Intrauterine mispositioning or constraint.
 - Oligohydramnios (low amniotic fluid).
 - Compartment syndrome in the intrauterine environment.
- Genetic/Developmental Factors:
 - Congenital anomalies (e.g. spina bifida, Arnold-Chiari syndrome).
 - Cervical spine abnormalities.
 - Absence or hypertrophy of cervical musculature.
- Other Associations:

1. Congenital hip dysplasia:

The association between CMT and DDH was established at a rate of 20% in 1972 by Hummer and MacEwen (38). Since then, other studies have explored this issue and refined the results. Von Heideken et al.(39) reviewed major studies on the co-existence rates of CMT and DDH, reporting rates ranging from 2% to 20% making it the most common associated anomaly. They then retrospectively studied 295 cases diagnosed with either CMT or DDH, finding a rate of 3.7% of DDH among patients referred for CMT. The incidence of CMT among patients referred for DDH was 5.9%. Among all patients with torticollis, 12.5% also had DDH, while 7.9% of children with dysplasia also had torticollis. Minihane et al. (40)report that DDH is ipsilateral to congenital torticollis in 75% of cases. This can be explained by intrauterine positioning or by the postural reaction caused by asymmetry of the head or hip. Cheng et al. (41) found that the incidence of DDH was correlated with the severity of CMT. Since these conditions are frequently associated, it is crucial to rely on evidence-based data to remain alert to the clinical signs necessary for early detection of these conditions.

In our study, we didn't note any patients with the 2 conditions associated.

2. Plagiocephaly:

It is defined as a cranial deformation usually presenting as unilateral flattening, is present in 80% to 90.1% of CMT cases (3). If the cranial deformation begins in utero, it can worsen if the child sleeps on their back and spends most of their awake time lying down, as they do not yet have the musculature needed to change their head position. Consequently, a significant increase in the incidence of CMT has been observed in the United States since 1992 and Canada in 1999, the year when the recommendation to lay infants on their backs to reduce the risk of sudden infant death syndrome was introduced. However, sleeping on the back is not the only cause of congenital torticollis, as this condition existed before this recommendation, and the majority of infants who sleep on their backs do not develop CMT.(42)

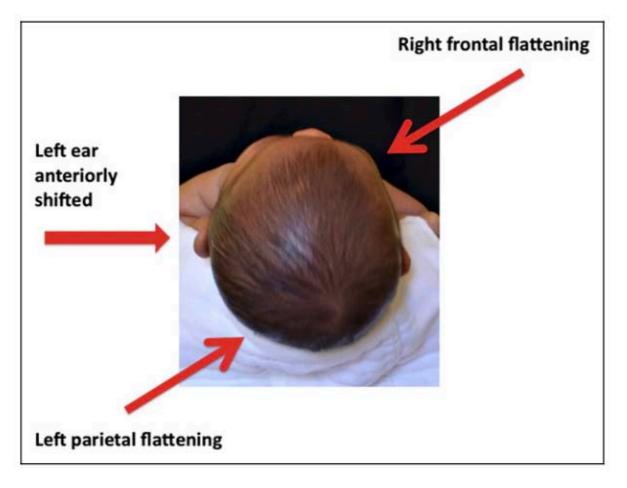


Figure 56 : Features of asymmetry related to plagiocephaly in a 4-month-old infant.(3)

3. Foot deformities(Clubfoot varus equinus):

Clubfoot is a congenital foot deformity partly related to intrauterine malpositioning.

4. Scoliosis:

Few studies have examined the relationship between idiopathic infantile scoliosis and CMT. According to Hamanishi (43), the primary cause of asymmetry in children is often a tight uterine environment. It is logical to link these two conditions because intrauterine mispositioning that leads to CMT places the cervical spine in an asymmetric position.

Consequently, the rest of the spine and limbs are likely also in restricted asymmetric positions. This connection helps explain the association between clubfoot, hip dysplasia, congenital plagiocephaly, and CMT. Ballock's study(44) indicates that scoliosis can also cause torticollis, though it usually presents as non-muscular torticollis. In Wynne-Davies' study(28) , plagiocephaly was present in all 97 cases of scoliosis examined, with cranial flattening always occurring on the side of the scoliotic curve's convexity.

→ While birth trauma was historically considered the main cause, more recent evidence suggests intrauterine factors may play a larger role in the etiopathogenesis of CMT. However, the exact underlying mechanisms are still not fully understood (5)(7)(34)(45).

IX. <u>Diagnosis :</u>

1. Anamnesis:

The diagnosis of CMT is primarily clinical based on the characteristic head tilt, limited neck range of motion, and palpable mass but a detailed history is crucial to confirm the congenital nature of the condition, rule out other causes, and guide management . Early recognition through careful history-taking is important, as delayed diagnosis can lead to more severe craniofacial asymmetry and the need for surgical treatment (46).

The anamnesis should look for the following key points :

- <u>Pregnancy and delivery :</u>
 - Parity : Understanding the parity status of mothers can provide insights into potential factors influencing the development of congenital conditions such as wryneck and may guide further research into prenatal care and maternal health interventions.

CMT is relatively often observed in children of primiparas. Ho et al. noted an incidence of 53% in children with CMT from primiparous mothers , and there was a high occurrence of

traumatic childbirth.(5)

In our study, 76.5% of the mothers were primiparous with a poor pregnancy follow-up which indicates an inadequate access to or utilization of prenatal care services, emphasizing the importance of improving prenatal care services to optimize maternal and fetal health outcomes.

- Multiple pregnancy: The heightened susceptibility to CMT in multiple pregnancies is likely attributed to the constrained intrauterine environment, particularly in the later stages of gestation. This can lead to atypical fetal positioning and an increased likelihood of birth trauma affecting the sternocleidomastoid muscle. Timely identification and management of CMT are crucial to mitigate potential complications such as craniofacial asymmetry in these vulnerable infants. In essence, multiple gestation stands out as a recognized risk factor for CMT, with the more severely impacted and lower positioned twin facing the greatest susceptibility. Therefore, vigilant monitoring for CMT in cases of multiple pregnancies is essential to provide valuable insights into the potential factors influencing fetal development and the associated healthcare needs for both mothers and infants.(34)(47)

In our study only 2 patients out of 17 were issued from multiple pregnancies.

- Birth weight: There appears to be a relationship between larger birth weight and an increased risk of CMT, particularly in male infants:

The study by Płomiński et al. found that male infants with CMT had a significantly greater birth weight (3632 g) compared to females (3299 g). The same study also reported that CMT was more common in boys (54%) than girls (46%), which the authors hypothesized may be related to the greater birth weights seen in male infants .(5)

Unfortunately, in our study we couldn't get enough data about the birth weight of our patients. However, the study by Hardgrib et al. concluded that complicated birth or birth

trauma, which can be associated with macrosomia, may not be the main cause of CMT. Most children in their study were delivered at term without signs of birth complications or trauma.

So while larger birth weight, especially in males, appears to be associated with a higher incidence of CMT, the evidence is not conclusive that birth weight is a direct causative factor. (7)

- Gestational diabetes : Gestational diabetes can have significant implications for maternal and fetal health, including an increased risk of complications during pregnancy and childbirth, such as macrosomia that can lead into an elevated risk of traumatic birth which can increase the occurrence of congenital torticollis . However, according to Hardgrib et al even though gestational diabetes is associated with increased frequency of large for gestational age infants and birth injuries in general , the search results do not directly link it to an increased risk of congenital muscular torticollis. The etiology of CMT appears to be multifactorial, with intrauterine and prenatal factors potentially playing a more important role than birth trauma.(7)

In our study, 5 patients of 17 suffered from gestational diabetes.

- Term : The findings from the studies by Płomiński et al. and Hardgrib et al. suggest that pregnancy term does not appear to be a significant factor in the development of CMT. Both studies reported that the majority of children diagnosed with CMT were delivered at term without any signs of birth complications or trauma.(7)(5) (which aligns with our study where 94.1% of the patients were delivered at full term.)

Płomiński et al.'s study indicates that being delivered at term does not seem to contribute to the occurrence of CMT. Similarly, Hardgrib et al. found that complicated births or birth trauma may not be the primary cause of CMT, pointing towards intrauterine and prenatal factors as potential contributors to the development of this condition.

Based on the information provided in these studies, there does not seem to be a strong association between pregnancy term and the incidence of congenital torticollis. Infants born at term, without experiencing any birth complications or trauma, were still diagnosed with CMT.

> Birth experience (labor, position and use of instruments) : CMT may develop following a difficult or prolonged labor, particularly when the infant is delivered in a breech position or is significantly larger in size. During such challenging births, the sternocleidomastoid muscle can be subjected to excessive stretching pulling forces or use of instruments such as vacuum or forceps, potentially leading to muscle tears. This muscle injury can subsequently trigger fibrosis and shortening of the affected muscle, ultimately causing the characteristic twisting of the neck observed in CMT patients. (46)(34)

Furthermore, the study by Płomiński et al. corroborates this association, finding that congenital torticollis is more likely to occur following difficult births involving large infants or breech deliveries. The researchers attribute this increased risk to the muscle injury and subsequent fibrosis that can result from these challenging delivery scenarios(5).

In summary, a long and complicated labor, especially when complicated by factors such as fetal macrosomia or breech presentation, can significantly increase the risk of congenital muscular torticollis. The potential for muscle injury, fibrosis, and subsequent shortening of the sternocleidomastoid muscle during these challenging deliveries appears to be the underlying mechanism linking difficult births to the development of CMT.

The study conducted by Hardgrib et al. in 2017, as published in the Journal of Orthopaedics and Traumatology, challenges the notion that complicated birth or birth trauma is the primary cause of CMT. Their research revealed that most children diagnosed with CMT were born at term without any indications of birth complications or trauma. Additionally, the study found no significant differences in birth characteristics between children with left- and rightsided torticollis, between genders, or between those managed conservatively versus those requiring surgery. These findings led the authors to suggest that factors related to intrauterine and prenatal conditions may play a more crucial role in the development of CMT than birth trauma or complications. They emphasized the necessity for further exploration of potential intrauterine risk factors associated with CMT.(7)

In contrast, a study by Kim et al. identified a notable 16.6% of CMT patients presenting in the breech position, a significantly higher rate compared to the general population's 3-4%.(48) The authors of this study proposed that being in the breech position during fetal development could have a substantial impact on the shortening and fibrosis of the sternocleidomastoid muscle, potentially contributing to the development of CMT.(32)

This goes along with our study where 58.8% of the patients issued from a complicated and prolonged labor, 29.4% had a traumatic birth, 52.9% were in a breech position and 35.3% were delivered with the use of instruments.

- When the head tilt was first noticed by the parents and how it has progressed over time.
- The presence of a mass or lump in the neck muscle that may have been present at birth.
- Difficulty feeding or turning the head to one side.
- Any associated developmental DDH, as CMT and DDH can coexist in up to 20% of cases.

2. <u>Clinical examination :</u>

CMT typically presents with a unilateral tilt of the head and neck, along with rotation, plagiocephaly, and sometimes a palpable fibrous mass. Diagnosis is usually made through a clinical examination that assesses both active and passive range of motion of the neck in rotation and tilt, and involves palpation of the SCM. A diagnosis is confirmed when there is a difference of more than 15° in range of motion between the two sides.(49)

The complexity of the SCM and the challenge of observing the maximum neck range of motion in newborns often result in underestimating the number of congenital torticollis cases, according to Stellwagen (49). Additionally, the values considered normal or excellent for children are often inappropriate, with rotation ranges from 75° to 120° and tilt ranges from 40° to 90°. Research has shown that for infants under one year old, the average cervical tilt is between 65° and 75°, and rotation is between 100° and 120°(49)(50). Restrictions in tilt are more easily detected than in rotation, so a child may have torticollis but still manage to bring their chin to shoulder level (90°) in rotation(49).

Clinical diagnosis is rarely made before 3 months of age. This can be attributed to several factors: facial asymmetry, which typically concerns parents and prompts them to seek medical advice, seldom appears before 3 months; additionally, midline head control usually begins around 3 months, making the asymmetry in head rotation and tilt more noticeable to parents. (51)

A physical examination alone is often insufficient for diagnosis, especially when head tilt is minimal or absent, and the SCM mass is difficult to palpate.(52)

Poole [28] describes asymmetric deformations of the face and cervical spine as the cranio-faciocervical scoliosis complex. Often, a scoliotic posture or a simple shoulder asymmetry is observed. These are signs of the spine adapting to the underlying deformation.(53)

In our study, All of the patients presented a head tilt of the affected side alongside with a limited head rotation of which 56,3% came with a $>15^{\circ}$ limitation and 43.8% with a $\leq 15^{\circ}$ limitation, only 23.5% exhibited a facial asymmetry, 47.1% a shoulder elevation of the opposite side, 17.6% presented a fibrotic mass, 11.8% a flat foot, 35.3% scoliosis and none of the patients presented a plagiocephaly or a hip anomaly.

3. Paraclinical examination :

The positive diagnosis of CMT is made clinically, with no supplementary tests capable of confirming it. However, when the diagnosis is challenging and differential diagnoses are being considered, paraclinical investigations and their negative results can be very helpful.

3.1. <u>Ultrasonography :</u>

This technique confirms that the palpable mass is indeed located within the SCM muscle and does not penetrate deeper tissues. By visualizing changes in the echo structure of the muscle, ultrasound can help rule out other diagnoses in cases of clinical uncertainty, such as:

- A paramedian thyroid mass.
- An embryonic cyst.
- Cervical lymphadenopathy: characterized by a variable echo structure, located anteriorly, and clearly separated from the SCM.
- Fibromatosis colli: This condition is a type of fibromatosis in infants that specifically affects the SCM, leading to fibrous changes that can form a palpable tumor, usually resolving spontaneously within a few months.
- Ultrasound imaging shows tissue that is more echogenic than the surrounding muscle, with a denser echo structure, situated within the muscle. Other ultrasound findings, using the same equipment, have demonstrated a well-defined mass within the muscle that is uniformly echogenic but hypoechoic compared to the surrounding muscle. During real-time examination, the mass moves synchronously with the SCM muscle (52)(49)(54)(55).

In our study, only one of 17 patients benefited of an ultrasound examination showing a fibrotic olive.

3.2. Standard X-rays:

While it's generally advisable to avoid X-rays in newborns unless absolutely necessary to minimize radiation exposure at their tender age, radiographic examinations serve a dual purpose. It's crucial for all infants with congenital muscular torticollis to undergo a thorough evaluation to detect any potential complications or associated abnormalities.

This radiological assessment typically involves obtaining X-rays of the cervical spine (frontal and lateral views), as well as of the thoracic and possibly lumbar spine (frontal and lateral views). Additionally, frontal pelvic X-rays and focused hip images are essential for identifying any spinal curvature anomalies and potential congenital hip dislocations, given their relatively high occurrence.

Furthermore, radiological examinations play a crucial role in cases where the underlying diagnosis is challenging, particularly when there are concerns about traumatic, malformative, or infectious origins.(56)(57)

In our study, 52,9% of the patients underwent Standard X-rays examination that showed a deviation of the cervical spine to the affected side.

3.3. CT-scan/MRI:

While CT and MRI offer the advantage of identifying structural changes in the brain, cervical spine, and soft tissues, they have limitations in detecting small amounts of fibrosis or masses within various muscles, as seen in congenital muscular torticollis. Additionally, both CT and MRI procedures are costly, time-consuming, and require general anesthesia(52) (58)(59). According to Parikh et al.'s study (59), MRI only identified 30% of cases of congenital muscular torticollis. Given that the risks outweigh the benefits, these imaging techniques are sparingly used for congenital muscular torticollis(59).

In our study, only 2 children of 17 benefited from a CT-scan which showed a high cervical scoliosis with convexity of the affected side and straightening of the cervical spine. and 1 from an MRI which showed a D12-L1 block vertebra.

X. Differential diagnosis :

It is essential to rule out other pathologies. Some of the potential differential diagnoses for congenital torticollis include (60)(61)(62):

- Vertebral anomalies like hemivertebrae and Klippel-Feil Syndrome.
- Unilateral congenital absence of the SCM muscle.
- Congenital scoliosis.
- Ocular torticollis.
- Sandifer syndrome.
- Arnold Chiari malformation.
- Neurological diseases.
- Visual disturbances.
- Syringomyelia.
- Cervical spine tumor.
- Brain tumor.

XI. <u>Treatment</u> :

1. Purposes :

Literature presents numerous therapeutic recommendations for a proper CMT management (54)(63)(64) :

- Therapeutic abstention with regular observation.
- The use of an orthosis combined with an active home stimulation program.
- Stretching or gentle manipulations, passive stretches, and a wide variety of surgical procedures.

Despite the many existing approaches, authors agree on various common therapeutic goals (27)(65):

- Ensuring there is no risk of vascular or nerve injury.
- Achieving a good functional outcome where the cheek can touch the shoulder on the opposite side of the contraction (a criterion for recovery).
- Minimizing aesthetic impact by reconstructing a symmetrical muscle contour in the neck and keeping scarring to a minimum.

2. <u>Means :</u>

2.1. Advice and Home Postures :

When a baby is born, their neck is short and weak, making traditional physiotherapy manipulations unsuitable. Torticollis often improves on its own at this age. However, it's important for parents to follow certain guidelines for bed positioning and stimulation activities :

- Avoid placing the baby on their stomach, as this can reinforce the incorrect head position and worsen the condition. Instead, place the baby on their back with their head turned to the opposite side of the torticollis.
- Position the crib so that the light encourages the baby to turn their head in the direction that corrects the torticollis.
- The mother should always interact and stimulate the baby from the corrective side, and toys should be placed on that side as well.

These tips should be combined with gentle movements to help the baby turn their head towards the affected side.(24)

2.2. <u>Physiotherapy :</u>

Studies indicate that manual stretching remains the most common treatment for CMT, both with and without tumors (65)(68)(69). The exact mechanisms of stretching in these cases are not fully understood (68). Research by Tang et al. (67)suggests that myoblasts within the tumor are crucial for its maturation and resolution, likely by producing normal myofibrils. With

the right environment and stimulation, these myoblasts can aid in the regeneration and repair of the affected sternocleidomastoid (SCM) muscle. Manual stretching can provide the necessary stimulation for muscle development.

Without this beneficial stimulation or in cases of severe damage, fibroblasts may take over, leading to progressive fibrosis, as observed in typical cases of late-stage torticollis. Additionally, Cameron et al. (70)found that a regular stretching program helps maintain the length of immature scar tissue in the fibrotic SCM during the remodeling phase and prevents cervical fascia contractures.

Unfortunately, most articles fail to detail their stretching parameters and do not adhere to a standardized manual stretching protocol. So far, according to our research, no study has compared the various parameters to establish the most effective protocol. All stretching parameters appear to be effective and are not questioned because they consistently produce good results.

| Series | Country | SCM Stretching Protocol | Results |
|--------------------------|-----------|---|--|
| Cheng et al. (68) | Hong Kong | Frequency: 3 times per week. Repetitions: 3 sets of 15. repetitions. Performed by: Physiotherapist. | Effective and safe in 95% of cases. |
| Celayir et al. (71) | Korea | Exercises: Flexion/Extension Rotation Lateral Flexion Repetitions: 10 repetitions each. Frequency: 8 times per day. Performed by: Both parents. | 100% success rate. No surgery required. |
| Demirbilek et al.(69) | Turkey | Repetitions: At least 40 repetitions. Frequency: 4-5 times per day. Performed by: Both parents. | Success rate depends on the starting age of treatment: • <3 months: No surgery required • 3-6 months: 25% surgery rate • 6-18 months: 70% surgery rate • >2 years: 100% surgery rate |

Table VI: Comparison of pre-operative physiotherapy outcomes.

2.3. Botulinum Toxin :

Botox was proven effective in treating torticollis cases resistant to standard treatments(58). In a study by Joyce et al.(72), 14 patients with persistent torticollis received Botox injections and were monitored for 22 months. Only one patient had a residual deficit of 10° and required surgery. This study suggests that Botox could play a valuable role in managing congenital torticollis by offering a 3 to 6-month window to improve range of motion and solidify progress. However, there is limited research on the long-term effects of Botox treatment for torticollis. It's worth noting that Botox administration carries risks, including the potential for hematoma or systemic diffusion, as it is administered under general anesthesia (72)(73).

2.4. Surgery:

A range of surgical techniques is available for treating torticollis, from simple tenotomy to muscle resection, including tenotomies at both ends and Z-lengthening plasty. These procedures aim to achieve complete tenotomy of the SCM muscle at various levels and are performed openly to ensure precise control over muscle sectioning, hemostasis, and spinal nerve preservation. Incomplete tenotomies increase the risk of recurrence. (63)(64)

The common goal is to restore symmetrical and full range of motion while maintaining a harmonious aesthetic appearance, particularly the muscular contour at the base of the neck, creating a symmetrical "V" shape.(74)

a. Unipolar tenotomy :

The low tenotomy should be deep and primarily posterior to ensure the sternal head is adequately addressed; any incision over the clavicular prominence risks unsightly scarring, hence it's advisable to perform the skin incision a finger's breadth above the bony prominence of the clavicle.

Early complications such as hematoma, skin dehiscence, or infection are rare, with

excellent long-term functional outcomes. However, it's important to note that fibrous bands recur on the clavicular portion in almost half of the patients, though without functional or aesthetic consequences. While loss of muscular contour is observed in 8 out of 10 cases, it generally isn't perceived as an aesthetic issue, unlike reported problems with unsightly scars, in 2 to 32% of the cases particularly when the scar is directly over the clavicle.(75)(76)

Advantages of this technique are :

- Less Invasive: Unipolar tenotomy is a less invasive procedure compared to bipolar tenotomy, which involves releasing both the sternal and clavicular heads of the SCM muscle.(77)(78)
- Faster Recovery: Unipolar tenotomy typically results in a faster recovery compared to bipolar tenotomy, as it involves a single incision and less tissue disruption.(77)(78)
- Lower Risk of Complications: Unipolar tenotomy has a lower risk of complications compared to bipolar tenotomy, such as nerve damage and bleeding, still a risk of clavicular head reattachment and formation of a lateral band, requiring secondary surgery remains possible.(77)(78)
- Improved Range of Motion: Unipolar tenotomy can improve the range of motion in children with congenital torticollis, allowing for better head movement and reduced stiffness.(77)(78)
- Effective in Younger Children: Unipolar tenotomy is effective in treating congenital torticollis in younger children, as it can be performed under general anesthesia and is less traumatic compared to bipolar tenotomy.(77)(78)

Disadvantages of this techniques are :

• Limited Release: Unipolar tenotomy only releases the sternal head of the sternocleidomastoid muscle, which may not be sufficient to fully correct the

deformity in some cases.(77)(78)

- **Risk of Recurrence**: Unipolar tenotomy carries a risk of recurrence of 26.5%, as the clavicular head of the SCM muscle may not be fully released.(77)(78)
- Limited Effectiveness in Older Children: Unipolar tenotomy may not be as effective in treating congenital torticollis in older children, as the muscle may have become more fibrotic and less responsive to the procedure.(77)(78)
- **Risk of Asymmetry**: Unipolar tenotomy may not fully correct facial asymmetry, which can persist even after the procedure.(77)(78)
- Limited Long-Term Follow-Up: There is limited long-term follow-up data available for unipolar tenotomy in children with congenital torticollis, making it difficult to assess its long-term effectiveness.(77)(78)

b. Bipolar tenotomy :

Described by Barcat(79). This technique is applicable at any age, including adulthood or in cases of recurrence, but the ideal age is between 3 and 5 years, requiring some postoperative cooperation (80). At this age, recurrence rates are low, and cosmetic outcomes are favorable, especially when muscle continuity is preserved.

However, nerve complications due to stretching during immobilization have been reported (80):brachial plexus paralysis, deltoid paralysis, although spontaneous recovery is common.

Advantages of this technique :

- More Complete Release: Bipolar tenotomy involves releasing both the sternal and clavicular heads of the SCM muscle, providing a more complete release of the muscle and potentially better outcomes.(81)
- Improved Range of Motion: Bipolar tenotomy can improve the range of motion in children with congenital torticollis, allowing for better head movement and reduced stiffness.(81)

- **Reduced Recurrence**: Bipolar tenotomy may reduce the risk of recurrence(11%) compared to unipolar tenotomy, as it releases both heads of the sternocleidomastoid muscle.(81)
- Better Facial Asymmetry Correction: Bipolar tenotomy can improve facial asymmetry, which is often associated with congenital torticollis.(81)
- Effective in Older Children: Bipolar tenotomy can be effective in treating congenital torticollis in older children, as it provides a more complete release of the muscle and can improve facial asymmetry.(81)

Disadvantages of this technique :

- **More Invasive**: Bipolar tenotomy is a more invasive procedure compared to unipolar tenotomy, involving two incisions and potentially more tissue disruption.(81)
- Higher Risk of Complications: Bipolar tenotomy carries a higher risk of complications compared to unipolar tenotomy, such as nerve damage and bleeding.(81)
- Longer Recovery Time: Bipolar tenotomy may require a longer recovery time compared to unipolar tenotomy, as it involves a more extensive procedure.(81)
- Higher Cost: Bipolar tenotomy may be more expensive compared to unipolar tenotomy, due to the additional incision and potential for more extensive tissue disruption.(81)

| Study | Country | Number of patients | Surgical technique | Scoring system | Results |
|----------------------|---------|--------------------|-----------------------|-------------------------|--|
| Seyhan et al.(82) | Turkey | 11 | Bipolar | Neck ROM | All had satisfactory ROM |
| Sudesh et al.(83) | India | 14 | Bipolar | Cheng and Tang score | Excellent: 3; Good: 7; Fair: 2; Poor :2 |
| Sahu et al.(84) | India | 34 | Bipolar/Unipolar | Cheng and Tang score | Excellent: 30; Good: 4 |
| Our study | Morocco | 17 | Bipolar/Unipolar | Cheng and Tang | Excellent: 4; Good: 12. |

Table VII: Comparison of Bipolar/Unipolar tenotomy outcomes.

c. Myotomy :

The muscle is accessed in its lower third through a transverse incision. Resection targets the area of retraction or fibrosis, although some have described complete muscle excisions. The correction is immediate but does not guarantee the prevention of distant adhesions, and the aesthetic result is subpar. This technique is typically reserved for older children, particularly in cases of recurrence.

d. Z-plasty:

This technique, pioneered by Jones in 1923 and later popularized in France by Barcat(79) and Dubousset (85). This technique offers a more natural solution with minimal recurrence rates. The aesthetic outcome is highly satisfactory, with a nearly invisible skin scar and preserved muscular contour and sternal "V". It's considered the technique of choice by many teams (85), as it allows for customizable elongation without the risk of unsightly scars, while maintaining symmetrical neck contouring.

Depending on the extent of retraction, immobilization with posture braces may not always be required, and gentle resumption of unrestricted movements can act as selfrehabilitation, yielding comparable results (85).

Advantages of Z-plasty:

- Improved Cosmetic Outcome: Z-plasty can help preserve the normal V-contour of the SCM muscle, leading to a better cosmetic outcome compared to other surgical techniques. This is particularly important in older children where the appearance of the neck is a greater concern.(85)
- Complementary to Other Surgical Techniques: Z-plasty is often used in combination with bipolar tenotomy to achieve better correction of the head tilt and rotation.(85)

Disadvantages of Z-plasty:

- Increased Complexity and Duration of Procedure: Z-plasty is an additional surgical step, increasing the complexity and duration of the overall procedure compared to tenotomy alone. This may lead to a higher risk of complications and a longer recovery period for the patient.(85)
- Limited Data on Outcomes: More research is needed to fully evaluate the longterm effectiveness and outcome of this technique.(85)
- Potential for Suboptimal Correction: While Z-plasty can improve the cosmetic outcome, it may not be as effective as bipolar tenotomy in fully correcting the head tilt and rotation in some cases.(85)

| Study | Number of the patients | Technique | Results |
|-------------------------------|------------------------|-----------|--|
| Ekici et al.(85) – Turkey– | 6 | Z-plasty | Excellent and good results were noted in all patients. |
| Our study | 1 | Z-plasty | Excellent |

Table VIII : Comparison of Z-plasty outcomes.

e. Endoscopy:

This technique, pioneered by ENT specialists, aligns with the current trend towards minimally invasive surgery. It involves a small 2 cm retro-auricular approach, perpendicular to the ear's long axis and hidden within the scalp near the mastoid. Precise subcutaneous dissection is crucial to identify the great auricular nerve. Once located, a traditional tenotomy is performed under endoscopic guidance, allowing visualization of spinal nerve branches during the procedure. This approach preserves deep muscle fibers, safeguarding the jugulo-carotid plane without compromising long-term release outcomes. Alternatively, a variant employs an axillary approach followed by a subcutaneous tunnel to selectively dissect and cut retracted fibers of the lower SCM and scalene muscles, controlling intraoperative mobility gain as needed. A foam cervical collar is worn for seven days, followed by approximately six weeks of passive and active mobilization.

These endoscopic release techniques offer the advantages of inconspicuous scarring, while maintaining both proximal and distal muscle innervation, thus preventing aesthetic issues in neck contours. Risks of neighboring nerve or vascular injuries are rare with proficient technical execution. (86)(87)(88)

- In our study, 82.4% of the patients underwent a unipolar tenotomy, 11.8% a bipolar tenotomy and 5.9% a Z-plasty.

f. Post-operative care : immobilization and physiotherapy :

Regardless of the surgical method employed, authors unanimously emphasize the significance of postoperative care, though they vary in their recommendations regarding orthopedic interventions. While some authors (89) advocate for immediate orthotic application in the days following surgery, others opt for a plaster collar. However, most recent publications suggest simple physiotherapy maneuvers during the initial two months, avoiding any form of support or orthosis. For older children, a simple foam cervical collar without fixed constraint is typically recommended.

Nevertheless, authors advocate for a straightforward yet continuous immobilization approach, using two sandbags immediately postoperatively, followed by the application of a basic collar from the third day onwards for a duration of six weeks. (90)

The postoperative immobilization strategy for congenital muscular torticollis remains a subject of debate .Various techniques are used, including traction, casting, halo vests, and collars. In the immediate postoperative period, patients often maintain their head in its preoperative position to alleviate pain, leading to poor compliance with prescribed exercises. This can result in the re-tightening of released structures if not addressed promptly. Administering gentle cervical traction immediately after surgery helps maintain correction. Once postoperative pain diminishes, transitioning to a collar and implementing an exercise regimen becomes feasible to achieve optimal outcomes. Ensuring strict adherence to the postoperative protocol through regular follow-up and patient motivation is crucial for achieving consistently positive results. Although the study did not demonstrate improved outcomes with cervical traction, we strongly advocate for its use along with strict adherence to physiotherapy protocols to assist patients in correcting abnormal head and neck positions postoperatively.(83)



Figure 57: Postoperative immobilization techniques:

Postoperative immobilization techniques: a Cervical collar used after three weeks postoperatively, b Head halter (seen from above). c Head halter (seen from the side). The head

halter gives continuous cervical traction for the first three weeks and night-time traction for the next three weeks.

- In our study, 47,1% of the patients were prescribed a plaster neck brace, 35,3% a regular neck brace, 11.8% a cervicothoracic cast and 5.9% a rigid neck brace, with a duration going from 4 days to 6 months with a discontinuous wear of 12hours/day. Also, all of our patient were prescribed a postoperative physiotherapy starting from 10 days to 30 days post-surgery, with an average duration of 3.82 months.

3. Indications :

The indications for treatment of torticollis vary based on the mode of presentation (with or without palpable swelling), age at onset, associated clinical signs, and the presence of complications such as facial asymmetry or plagiocephaly (98, 99). However, age at diagnosis is the most critical factor according to the literature.

- Newborns:

When the clinical presentation strongly suggests congenital torticollis, additional examinations like ultrasound, CT, or MRI are unnecessary. Immediate stretching is not required; positioning techniques administered by an experienced physiotherapist are sufficient (35)(54). If muscle tightness persists without visible tumors, gentle stretching or massages should be combined with positioning to restore mobility. Studies report a 70% resolution rate with home positioning, increasing to 99% with stretching(32) (91). Surgery is considered a last option for cases with:

- Diagnosis after 1 year of age.
- Persistent abnormal posture after 6 months of stretching.
- Passive lateral rotation deficit > 15 degrees.
- Severe craniofacial asymmetry.

• Poor responders to conventional treatment.

Poor responders often exhibit symptoms like cervical swelling during stretching, persistent muscle stiffness after 6 months, right-sided involvement, a history of obstetric difficulties, late torticollis diagnosis (over 12 months), and a cervical rotation limitation greater than 15° (71)(92)(93). Surgery is generally considered after 18 months to 2 years for French authors like Dubousset (64)7/26/2024 10:42:00 AM, while others recommend waiting until the age of reason (7–8 years) (94), with the best results seen after age six(95).

- Infants: 28days-23months

Postural torticollis in infants typically does not require specific treatment; some manipulations and home positioning help restore symmetric muscle tone. However, discovering torticollis at the fibrous retraction stage indicates a lesser likelihood of regression with conventional treatment and a greater need for surgical intervention(96)(97).

- Older Children: >2 years

Torticollis is increasingly diagnosed at ages two or three, and sometimes as late as adolescence or beyond. At this stage, stretching and manual methods are ineffective, making surgery the only viable option. However, the later the surgery, the less effective it is at correcting facial asymmetry (97) (98). Many authors now advocate delaying tenotomy indications, as over 90% of cases seen after age ten achieve good results (76). For others, like Minamitani (99)(100)(101), outcomes are favorable even after age seven, both aesthetically and functionally, although a visible scar at the base of the neck and minor residual inflection in one out of four cases may persist. Surgical techniques involve a mid-muscle section with Z-plasty, ensuring the incision follows a neck crease for aesthetic reasons.

For very late diagnoses, the primary challenge for practitioners is differential diagnosis and interpreting muscle retraction as a primary issue, without overlooking potential underlying bone, tumor, or ophthalmic abnormalities. This underscores the importance of thoroughly assessing bone, central, and peripheral neurological integrity in children over three years old seen for the first time.

XII. <u>Post-Operative Follow-Up Protocol</u>

1. Initial Follow-Up:

- **Timeframe**: 1-2 weeks post-operatively
- Assessment: Evaluate the patient's pain, swelling, and wound healing.
- Instructions: Provide guidance on wound care, pain management, and physical therapy.

2. Intermediate Follow-Up:

- **Timeframe**: 4-6 weeks post-operatively
- Assessment: Evaluate the patient's range of motion, strength, and any signs of complications.
- Instructions: Continue physical therapy and provide guidance on maintaining proper posture and movement.

3. Final Follow-Up:

- **Timeframe:** 3-6 months post-operatively
- **Assessment**: Evaluate the patient's final range of motion, strength, and any residual symptoms.
- Instructions: Provide guidance on maintaining proper posture and movement, and schedule follow-up appointments as needed.

XIII. Complications :

- 1. <u>Short–Term</u> :(Within the first month post–surgery):
 - Infection: Signs include redness, swelling, warmth, pain, and discharge at the surgical site.
 - Hematoma: Accumulation of blood at the surgical site, causing swelling and discomfort.
 - Wound Dehiscence: Partial or complete separation of the surgical incision.
 - **Pain and Discomfort**: Post-operative pain that may require management with medication.
 - **Poor Wound Healing**: Delayed healing due to infection, poor nutrition, or other underlying conditions.
 - Adverse Reaction to Anesthesia: Nausea, vomiting, or more severe reactions in rare cases.
 - Suture Loosening: Early loosening of sutures leading to wound dehiscence or delayed healing.
- 2. <u>Mid-Term</u> :(1 to 6 months post-surgery):
 - Scar Formation: Hypertrophic or keloid scars may form, causing cosmetic concerns or discomfort particularly if it is placed too low and horizontally over the clavicular insertion of the SCM muscle, which should be avoided during incision planning. Incomplete muscle sectioning poses the most significant issues; frequently, only the sternal head is cut, leaving the clavicular head prominent, resulting in an unsightly bulge and a very visible suprasternal cavity.(58)(72)
 - Persistent fibrous bands after complete tenotomy are unpredictable and sometimes require secondary resection in 6 to 7% of cases, which usually results in excellent

outcomes. Fortunately, these fibrous bands, which appear 3 to 4 months after surgery, do not limit mobility or cause recurrence.

• Recurrence of Torticollis: Incomplete correction or recurrence of muscle tightness leading to a return of torticollis symptoms. Recurrences of torticollis following surgery are uncommon and are often linked to delayed treatment. These recurrences typically present as a tendency for the head to tilt, without significant limitation in rotation, often due to advanced dorsal kyphosis in late-stage congenital torticollis. Despite this, such issues are generally well tolerated and tend to improve over time.

True recurrences, however, involve progressive limitations in neck rotation appearing two to three months after tenotomy, which do not improve despite post-operative care. These true recurrences are more frequently associated with low, pre-clavicular incisions that are positioned too medially.(72)(58)

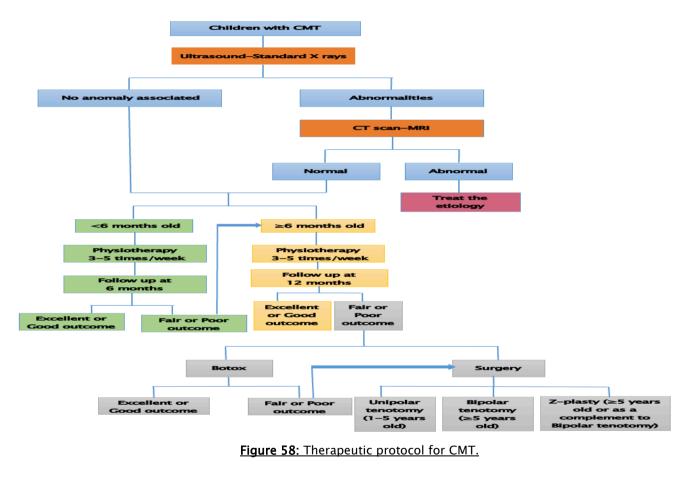
- Stiffness and Reduced Range of Motion: Inadequate physical therapy or excessive scar tissue can limit neck movement.
- Nerve Injury: Damage to nerves during surgery, leading to temporary or permanent weakness or sensory changes in the neck or shoulder area.
- Facial Asymmetry: While treatment leads to regression, it is never dramatic; postural or surgical correction of the head reduces the appearance of asymmetry, but much of the improvement occurs naturally. Complete regression in children operated on after their first year supports the case for early surgical intervention(102).
- **3.** <u>Long–Term</u> :(Beyond 6 months post–surgery):
 - Chronic Pain: Long-lasting pain in the neck or shoulder due to nerve damage, scar tissue, or muscle imbalance.
 - Functional Impairment: Persistent difficulty in neck movement affecting daily

activities and quality of life.

- **Cosmetic Concerns**: Visible scarring or residual asymmetry impacting the child's appearance.
- **Psychosocial Effects**: Potential impact on self-esteem and social interactions due to physical appearance or functional limitations.
- **Growth-Related Issues**: Potential for issues as the child grows, necessitating further surgical or therapeutic interventions.
- **Recurrence of Torticollis**: Return of symptoms over time, requiring additional treatment or surgery.
- Shoulder Asymmetry: Imbalance in shoulder height or positioning due to uneven muscle tension or surgical correction, in our case 2 patients exhibited a compensatory shoulder asymmetry.

Monitoring for these complications through regular follow-up visits and maintaining a proactive approach to management can help mitigate their impact and ensure the best possible outcomes for the child.

XIV. <u>Recommendations:</u>





Congenital muscular torticollis is a relatively uncommon condition characterized by the abnormal tightening of the SCM muscle due to fibrous tissue. The exact cause of this muscle tightening remains uncertain. Typically appearing in the early months of life, it results in a distinct neck posture where the head tilts to one side and rotates to the opposite side. Although some theories attribute it to trauma during childbirth, this remains a topic of debate.

It is often associated with congenital hip dislocation in medical literature, we did not find this correlation in our cases. The clinical presentation varies depending on when the condition is diagnosed.

In newborns, a palpable mass known as an "olive" within the SCM muscle is a characteristic feature that typically resolves within two to six months, leaving the muscle contracted. In older children, the neck deformity becomes more fixed, resulting in noticeable asymmetry in the positioning of the head and neck.

In advanced stages, congenital torticollis can lead to facial asymmetry or thoracic scoliosis due to compensatory postures. Before confirming a diagnosis of congenital torticollis, other potential causes such as postural issues, spinal malformations, congenital syndromes, trauma-related issues, infections, inflammations, or tumors should be ruled out.

Cervical ultrasound is the initial diagnostic tool of choice, offering valuable prognostic information. Further imaging studies such as X-rays, CT scans, or MRIs help assess the extent of spinal involvement and rule out other conditions.

The prognosis varies widely, with spontaneous recovery reported in 70–80% of cases, particularly with early initiation of rehabilitation efforts. Rehabilitation strategies initially focus on improving posture during activities like feeding and play. Active rehabilitation can be initiated as early as the second month of life, aiming to restore elasticity to the SCM muscle and to strengthen the opposing muscles.

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Surgical intervention should not be delayed, especially if there is significant muscle retraction beyond 12 months of age. Several surgical techniques are available, including SCM bipolar tenotomy for older children with pronounced retraction, Z-plasty for aesthetic preservation of the SCM contour, and low juxta-clavicular tenotomy using a minimally invasive approach. Following surgery, a neck brace is worn for four to six weeks to maintain proper posture, followed by ongoing rehabilitation which may start with a foam collar.

Despite potential concerns regarding appearance, especially in cases with later onset, the overall outlook for congenital torticollis is generally favorable.

Recurrence following surgical treatment often results from regrowth of fibrous tissue or inadequate initial muscle release.



<u>Identity:</u>

IP :

- Age:
- Sex :

ATCDS:

- Medical :
- Surgical :
- Family :

<u>Age of discovery :</u>

Pregnancy and childbirth:

Parity :

- Primipara 🗆
- Multipara 🗆
- Pregnancy follow-up : well followed
- Multiple pregnancy :yes \Box no \Box Gestational diabetes :yes \Box no \Box

Terme :

- Full 🗆
- Prematurity.
- Term overrun 🗆

Labour :

- Normal 🗆
- Complicated and prolonged \square

Position :

Birth :

- Normal 🗆
- Traumatic □

Delivery :

- Natural birth \Box
- Episiothomy □
- Ceasarean section \Box
- Use of instruments (section, forceps) \Box

Birthweight :

| Neonatal | rescusitation : yes \Box | no 🗆 |
|----------|----------------------------|------|
| | | |

Clinical examination :

Weight and height :

poorly followed□

| Cranial perimeter : | | |
|---|---------------------------|----------------------------------|
| Head tilt on affected side : | yes 🗆 | no 🗆 |
| Head rotation : | | |
| normale □ | | |
| • Limited 🗆 | | |
| • Limitation degree : | ≤ 15° □ | >15° 🗆 |
| Facial asymmetry : | yes 🗆 | no 🗆 |
| • Plagiocephaly : | yes 🗆 | no 🗆 |
| Shoulder elevation on contralateral side : | yes 🗆 | no 🗆 |
| Olive/SCM fibrotic mass: | present 🗆 | absent 🗆 |
| Hip examination : | Normal 🗆 | dysplasia of the hip \square |
| | | |
| | | |
| Foot deformities : | yes 🗆 | no 🗆 |
| Foot deformities : Scoliosis : | yes □ yes □ | no 🗆 no 🗆 |
| | , | |
| | , | |
| Scoliosis : | , | |
| Scoliosis : Paraclinical : | yes 🗆 | no 🗆 |
| Scoliosis : <u>Paraclinical :</u> Ultrasound : | yes 🗆 | no 🗆 |
| Scoliosis : <u>Paraclinical :</u> Ultrasound : • Results : | yes □ done □ | no 🗆 not done 🗆 |
| Scoliosis : Paraclinical : Ultrasound : • Results : Standard X-ray : | yes □ done □ | no 🗆 not done 🗆 |
| Scoliosis : Paraclinical : Ultrasound : • Results : Standard X-ray : • Results : | yes □ done □ done □ | no 🗆 not done 🗆 not done 🗆 |

<u>Traitement :</u>

Physiotherapy :

- Duration :
- Frequency :
- Period :
- Efficiency :
- Protocol :

Surgery :

- Duration:
- Position :
- Incision :
- Technique used :
- Per-operative complications :

Post -operative mangement :

Post-operative immobilization :

- Means :
- Duration :
- Physiotherapy :
- Resumption date :
- o Duration :
- Frequency :

Results :

- <u>Tanabe's assessment criteria</u> : Excellent \Box Good \Box Fair \Box Poor \Box
- \succ complaints : none \Box with \Box
- > limitation of ROM of the neck : none \Box mild \Box severe \Box
- \succ facial deformity : absent \Box mild \Box obvious and objective \Box
- <u>Cheng and Tang's score</u>: 17-21pts: excellent □ 12-16pts: good □ 7-11pts: fair □ <7pts: poor □
 - Rotation deficit : <5° (3pts) □ 6°-10° (2pts)□ 11°-15° (1pt)□ >15° (0pt)
 □
 - Side flexion deficit : <5° (3pts) □ 6°-10° (2pts) □ 11°-15° (1pt) □ >15° (0pt) □
 - Craniofacial asymmetry : none (3pts) □ mild (2pts) □ moderate (1pt) □ severe (0pt) □
 - Scar : none (3pts) □ mild (2pts) □ moderate (1pt) □ severe (0pt) □
 - Band : none (3pts) □ lateral (2pts) □ lateral, clavicular (1pt) □ clavicular, sternal(0pt) □
 - Head tilt : none (3pts) □ mild (2pts) □ moderate (1pt) □ severe (0pt) □
 - Subjective assessment (cosmetic and functional) : excellent (3pts) □ good (2pts)
 - \Box fair (1 pt) \Box poor (0 pt) \Box



<u>Abstract</u>

Congenital torticollis -wryneck- is a benign generally painless affection. It is considered the third congenital orthopedic anomaly after congenital hip dysplasia and clubfoot.

Our study is a retrospective series made of 17 cases of congenital torticollis collected at the pediatric trauma-orthopedics department of the Mohammed VI University Hospital Center of Marrakech over a period of 14 years.

The purpose of the study is to enhance the knowledge of congenital torticollis and understand the medical, surgical and post-surgical aspects of its management.

In our series, the age of surgery varies from 11 months to 11 years with the two thirds of the patients underwent surgery at the age of 6 and older.

The diagnosis is essentially clinical with a detailed anamnesis in order to point out the main risk factors and understand its nuanced physiopathology and a thorough clinical examination.

The diagnosis usually comes out in front of a sterno-cleido-mastoid muscle retraction leading to a head tilt to the affected side and a limited range of motion.

Paraclinical tests are usually prescribed to eliminate differential diagnosis and look for possible associated deformities.

The treatment of congenital torticollis is primarily conservative based on early physiotherapy, but unfortunately it can be inefficient especially if prescribed at an older age, in that case, the indication to surgery presents itself. In our case, all of our patient had to go through either an unipolar tenotomy, a bipolar tenotomy or a z-plasty for better results.

Surgery alone is not enough to achieve optimal results, it has to be followed with the pairing of a post-operative immobilization and a meticulous post-operative physiotherapy of an average period of 3 weeks and 3 months respectively. This association has lead into excellent and good results in our study.

In conclusion, optimal management of congenital torticollis requires a multidisciplinary approach involving doctors, physiotherapists, surgeons and the patient's surroundings.

<u>Résumé</u>

Le torticolis congénital est une affection bénigne et généralement indolore. Il est considéré comme la troisième anomalie orthopédique congénitale après la dysplasie congénitale de la hanche et le pied bot-varus equin.

Notre étude est une série rétrospective portant sur 17 cas de torticolis congénital colligés au service de traumatologie-orthopédie pédiatrique du Centre Hospitalier Universitaire Mohammed VI de Marrakech sur une période de 14 ans.

Le but de l'étude est d'approfondir les connaissances sur le torticolis congénital et de comprendre les aspects médicaux, chirurgicaux et post-chirurgicaux de sa prise en charge.

Dans notre série, l'âge de la chirurgie varie de 11 mois à 11 ans avec les deux tiers des patients opérés à l'âge de 6 ans et plus.

Le diagnostic est essentiellement clinique avec une anamnèse détaillée afin de mettre en évidence les principaux facteurs de risque et de comprendre sa physiopathologie nuancée et un examen clinique approfondi.

Le diagnostic est généralement posé devant une rétraction du muscle SCM entraînant une inclinaison de la tête du côté atteint et une limitation de l'amplitude des mouvements.

Le traitement du torticolis congénital est avant tout conservateur, basé sur une physiothérapie précoce, mais malheureusement elle peut être inefficace surtout si elle est prescrite à un âge avancé, dans ce cas, l'indication de la chirurgie se présente. Dans notre cas, tous nos patients ont dû subir soit une ténotomie unipolaire, soit une ténotomie bipolaire, soit une plastie en Z pour obtenir de meilleurs résultats.

La chirurgie seule n'est pas suffisante pour obtenir des résultats optimaux, elle doit être suivie d'une immobilisation post-opératoire et d'une physiothérapie post-opératoire méticuleuse d'une durée moyenne de 3 semaines et 3 mois respectivement. Cette association a permis d'obtenir d'excellents et de bons résultats dans notre étude.

En conclusion, la prise en charge optimale du torticolis congénital nécessite une approche multidisciplinaire impliquant les médecins, les kinésithérapeutes, les chirurgiens et l'entourage du patient.

ملخص

الصعر الخلقي هو حالة حميدة وغير مؤلمة بشكل عام. ويعتبر ثالث أكثر حالات الشذوذ العظمي الخلقي شيوعاً بعد خلل التنسج الخلقي في الورك وحنف القدم الخيلي.

در استنا عبارة عن سلسلة بأثر رجعي ل 7 [حالة من الصعر الخلقي تم جمعها في قسم جراحة العظام والكسور لدى الأطفال في المستشفى الجامعي محمد السادس بمر اكش على مدى 14 عامًا.

كان الهدف من هذه الدراسة هو زيادة معرفتنا بالصعر الخلقي وفهم الجوانب الطبية والجراحية وما بعد الجراحة في علاجه.

في سلسلتنا، تراوحت أعمار المرضى الذين خضعوا للجراحة من 11 شهرًا إلى 11 عامًا، مع إجراء ثلثي المرضى الذين خضعوا للجراحة في عمر 6 سنوات أو أكثر.

يكون التشخيص سريريًا بشكل أساسي، مع وجود تاريخ مرضي مفصل لتحديد عوامل الخطر الرئيسية وفهم الفيزيولوجيا المرضية الدقيقة، وفحص سريري شامل.

يتم التشخيص بشكل عام عندما يكون هناك تراجع في عضلة العضلة القترائية ، مما يؤدي إلى إمالة الرأس على الجانب المصاب ونطاق محدود من الحركة.

إن علاج الصعر الخلقي هو علاج تحفظي في المقام الأول، يعتمد على العلاج الطبيعي المبكر، ولكن للأسف يمكن أن يكون هذا العلاج غير فعال، خاصةً إذا تم وصفه في سن متقدمة، وفي هذه الحالة تظهر الحاجة إلى الجراحة. في حالتنا، كان على جميع مرضانا الخضوع إما لقطع الوتر أحادي القطب أو ثنائي القطب أو رأب على شكل حرف Zللحصول على نتائج أفضل.

لا تكفي الجراحة وحدها للحصول على أفضل النتائج؛ إذ يجب أن يتبعها تثبيت بعد الجراحة وعلاج طبيعي دقيق بعد الجراحة يستمر لمدة 3 أسابيع و3 أشهر في المتوسط على التوالي. أنتج هذا المزيج نتائج ممتازة وجيدة في دراستنا.

في الختام، تتطلب المعالجة المثلى للصعر الخلقي نهجاً متعدد التخصصات يشمل الأطباء وأخصائي العلاج الطبيعي والجر احين وعائلة المريض.



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