

2015 대한소아재활발달의학회 추계연수강좌  
11-21-2015

## 사경의 수술적 치료/비수술적치료



아주대학교병원 재활의학과  
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The Center for Torticollis · The Children's Rehabilitation Clinic

임신영, MD, Ph D  
아주의대 재활의학교실

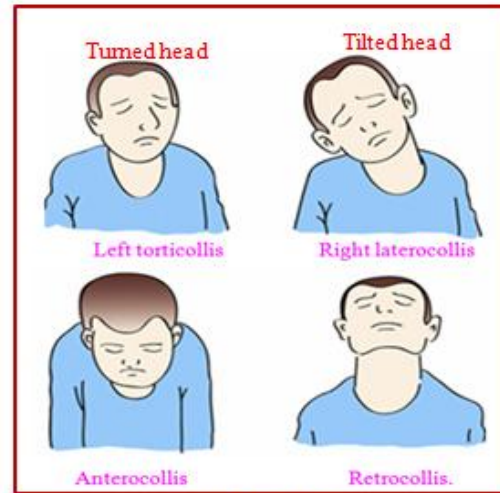
# Normal posture of the head and neck

Alignment of the head to vertical with the mouth horizontal

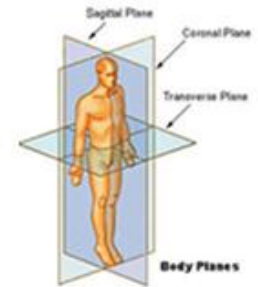


# 이상두위

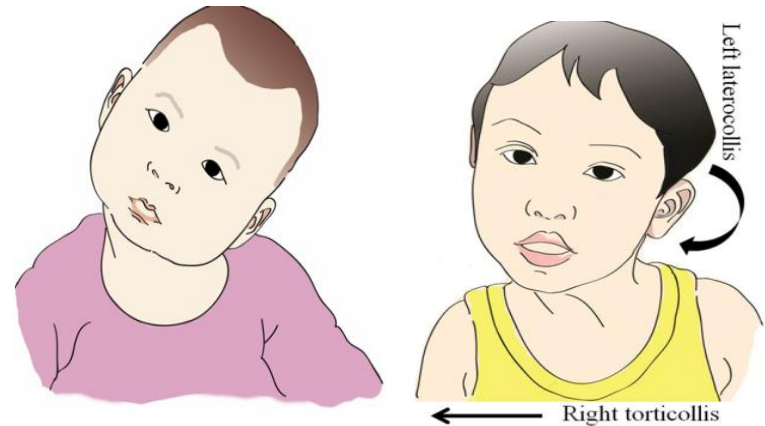
## Abnormal postures of the head and neck



TORTICOLLIS 사경  
 tortia (L. 'twisted')+collum (L. 'neck')  
 "wryneck", "twisted neck"



The Clinic for Torticollis



The Center for Torticollis

# 두경부이상자세/ 이상두위 Abnormal postures of the head and neck



TORTICOLLIS 사경

torta (L. 'twisted')+collum (L. 'neck')

“wryneck”, "twisted neck“



Left torticollis



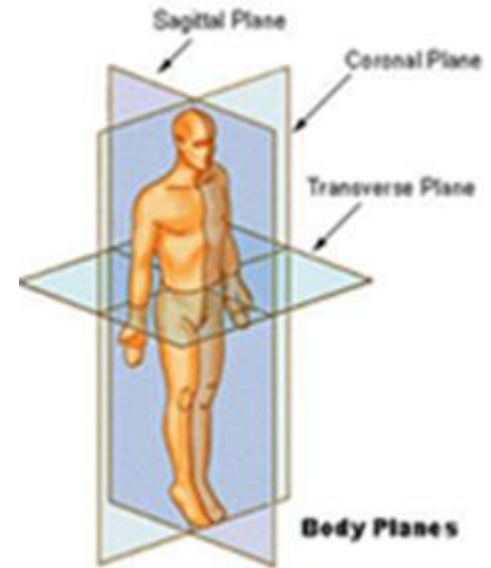
Right laterocollis



Anterocollis



Retrocollis.



Yim SY, Lee IY, Park MC, Kim JH. Differential diagnosis and management of abnormal posture of the head and neck. J Korean Med Assoc. 2009;52:726

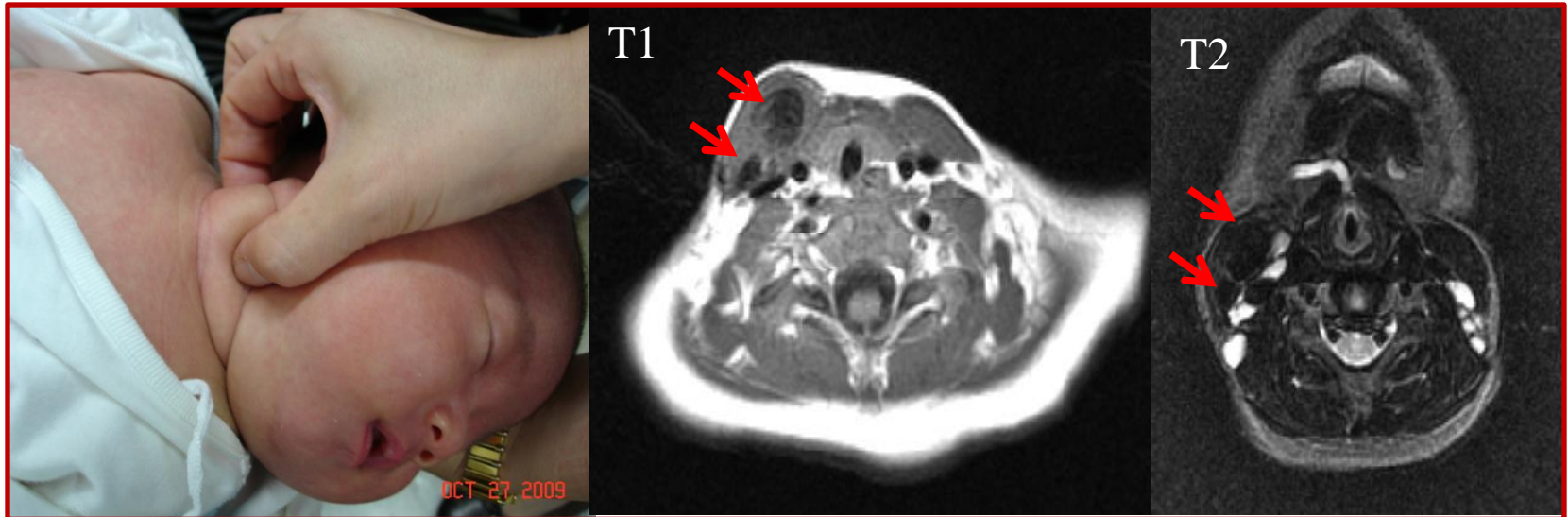
Kim JK, Yim SY. Clinical characteristics of abnormal postures of the head and neck caused by unilateral superior oblique palsy. J Korean Acad Rehabil Med. 2011;35:272

## Top 3 most common musculoskeletal problems in newborns

Clubfoot (congenital talipes equinovarus) 10%

Developmental dysplasia of hip: 0.6~7.6%

Congenital muscular torticollis 1.08~3.92%



- Dobbs MB, Gurnett CA. Update on clubfoot: etiology and treatment. Clin Orthop Relat Res. 2009;467:1146-53.
- Loder RT, Skopelja EN. The epidemiology and demographics of hip dysplasia. ISRN Orthop. 2011;2011:238607.
- Chen MM, et al. Predictive model for congenital muscular torticollis: analysis of 1021 infants with sonography. Arch Phys Med Rehabil 2005; 86: 2199-2203

1. 근성사경의 정의
2. 근성사경의 진단
3. 수술케이스와 비수술케이스의 감별
4. 근성사경의 자연경과

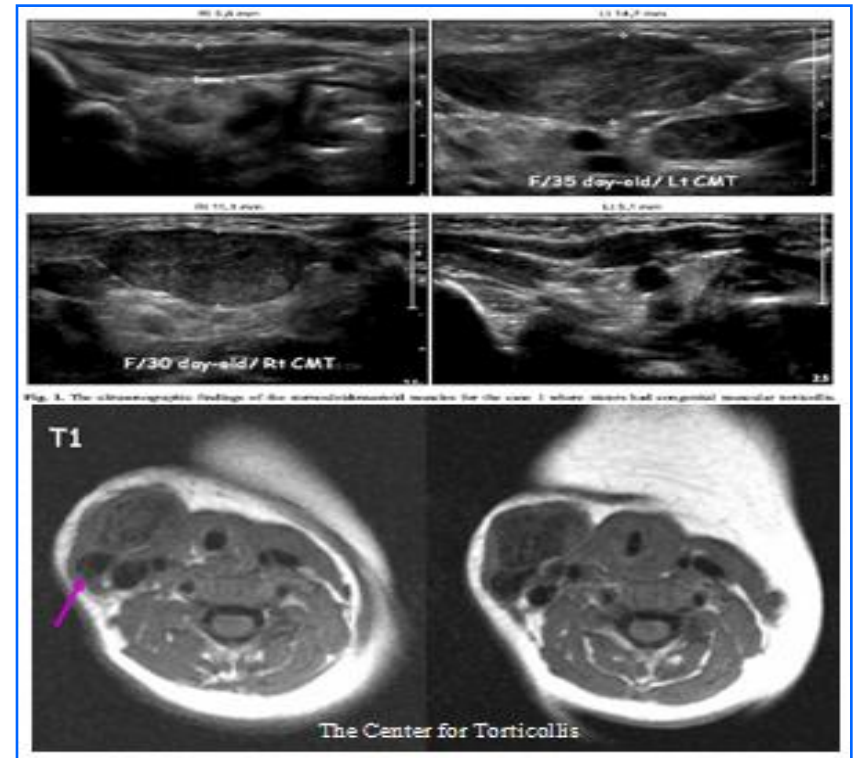


**아주대학교병원 사경센터**

The Center for Torticollis, Ajou University Hospital



## What is congenital muscular torticollis ?



**Unilateral palpable neck mass of muscle-consistency  
found exclusively in neonates**

Significant limitation of passive rotation of the neck toward the shoulder  
of the shortened sternocleidomastoid muscle (SCM) side

# What is congenital muscular torticollis ?



The Center for Torticollis

Unilateral palpable neck mass of muscle-consistency  
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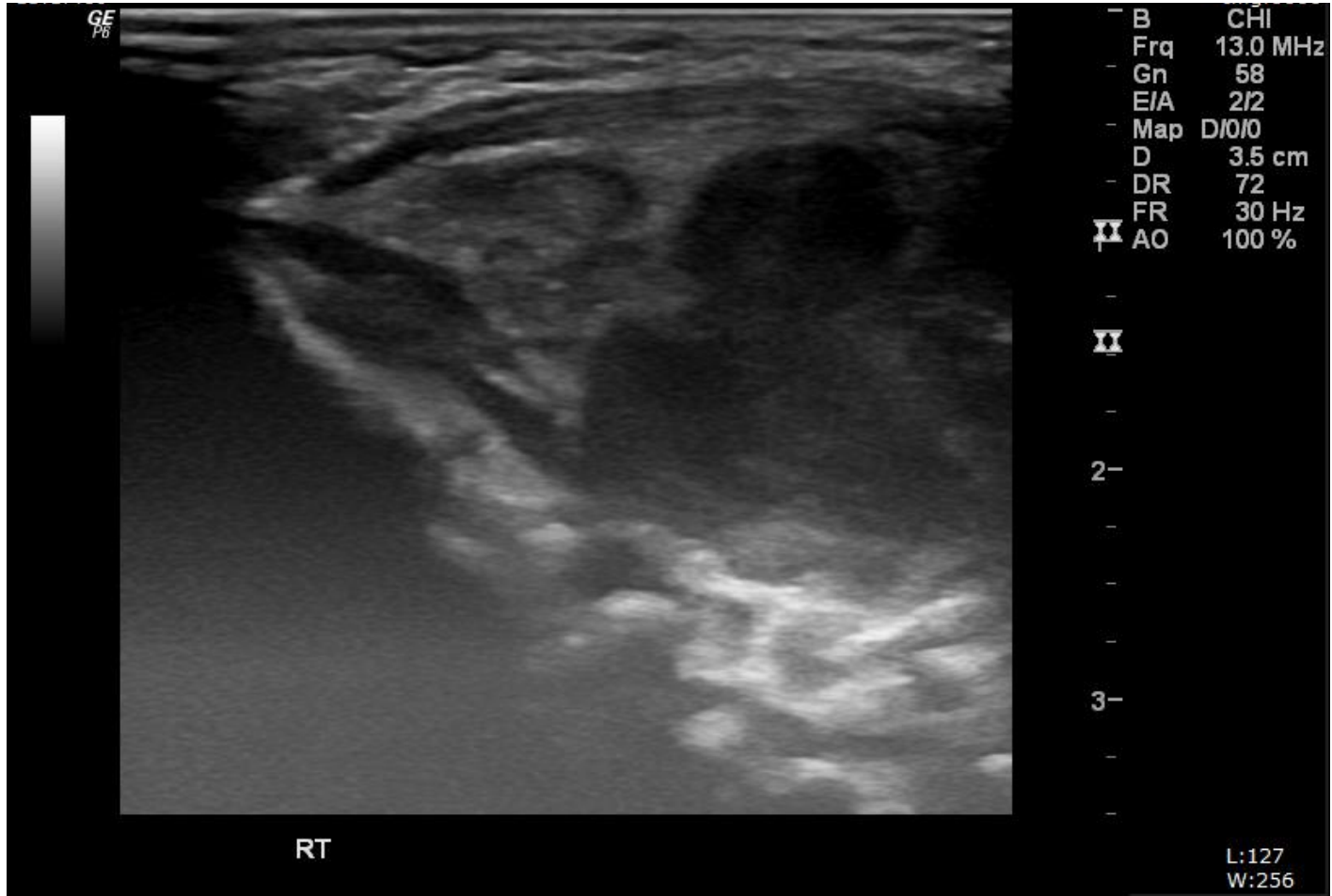
Significant limitation of passive rotation of the neck toward the shoulder  
of the shortened sternocleidomastoid muscle (SCM) side

Yim SY, Lee IY, Park MC, Kim JH. Differential diagnosis and management of abnormal posture of the head and neck.

J Korean Med Assoc. 2009; 52: 726

M/ 2mo

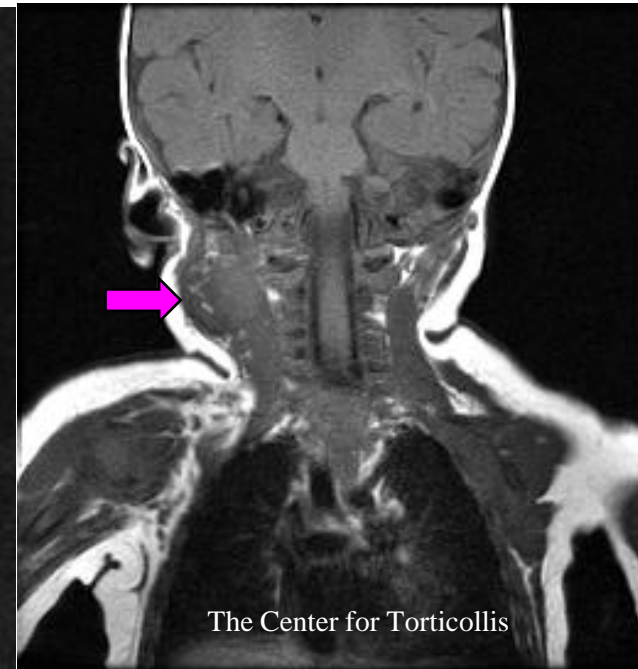
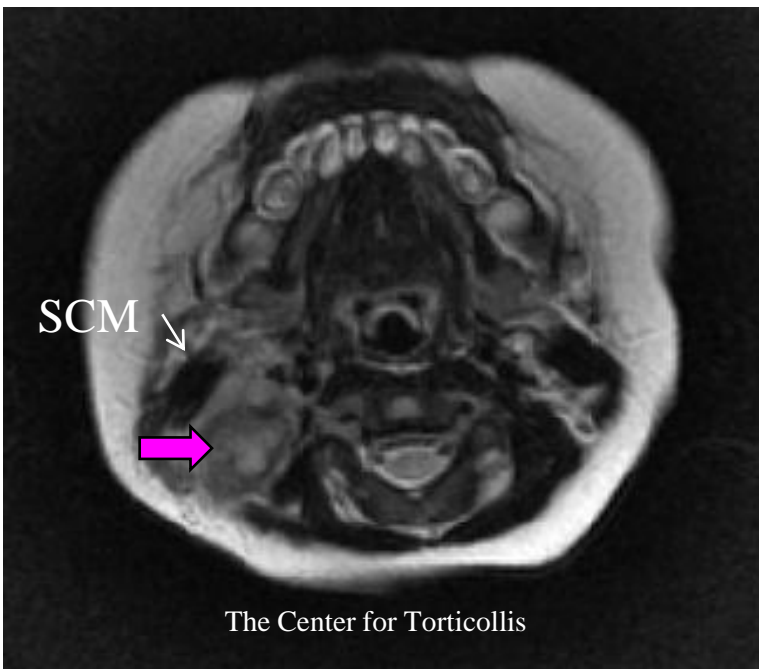
# Abscess in RSCM







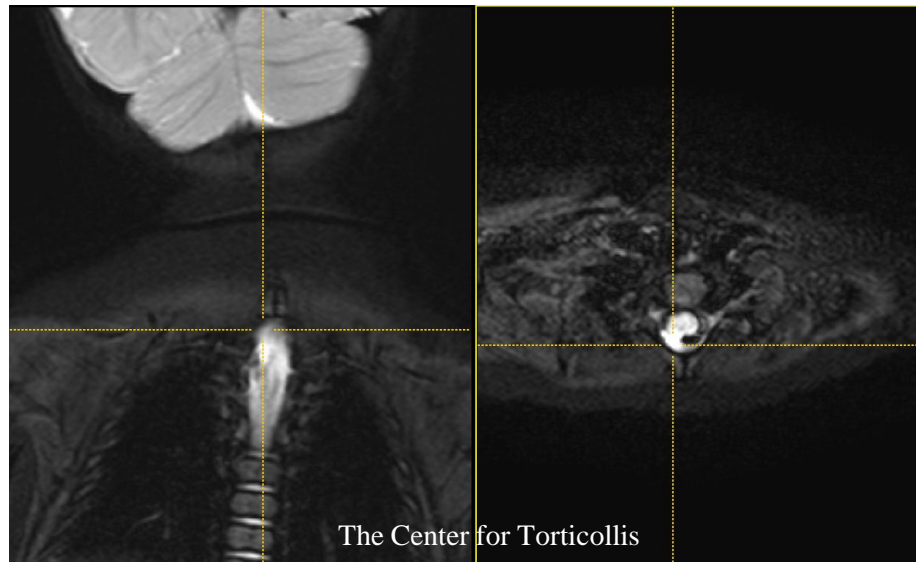
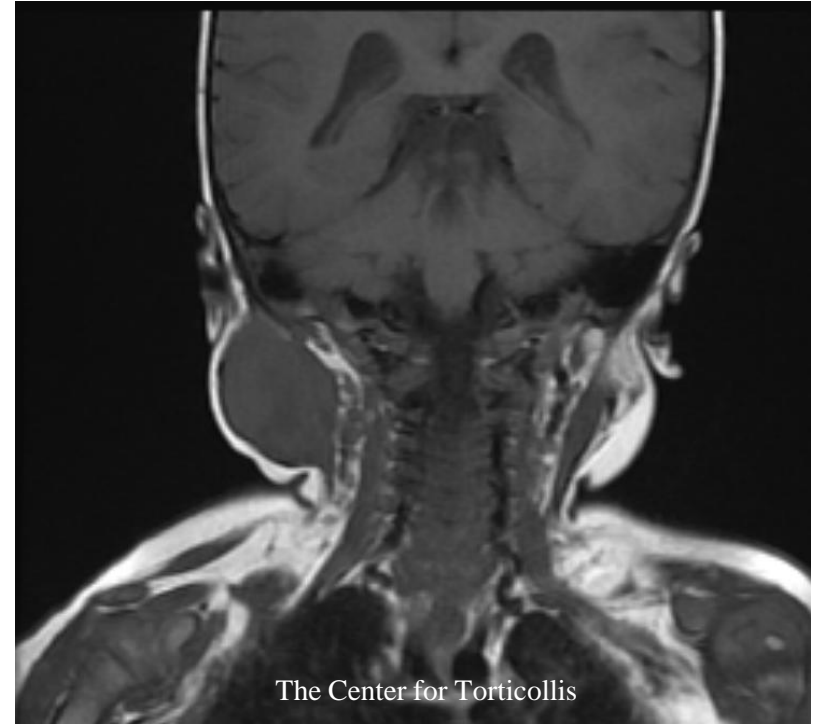
F/ 3 months old  
Acute torticollis  
secondary to deep neck abscess



Hwang JH, Lee HJ, Kim SO, Yim SY. Neck mass mimicking congenital muscular torticollis.  
Korean Academy of Rehabilitation Medicine, 2010: unpublished data

F/ 5mo

RCMT/ Lt DDH/ / Rt BPI/low grade fibromyxoid sarcoma



F/ 5mo

## RCMT/ Lt DDH/ / Rt BPI/low grade fibromyxoid sarcoma

### PATHOLOGICAL DIAGNOSIS

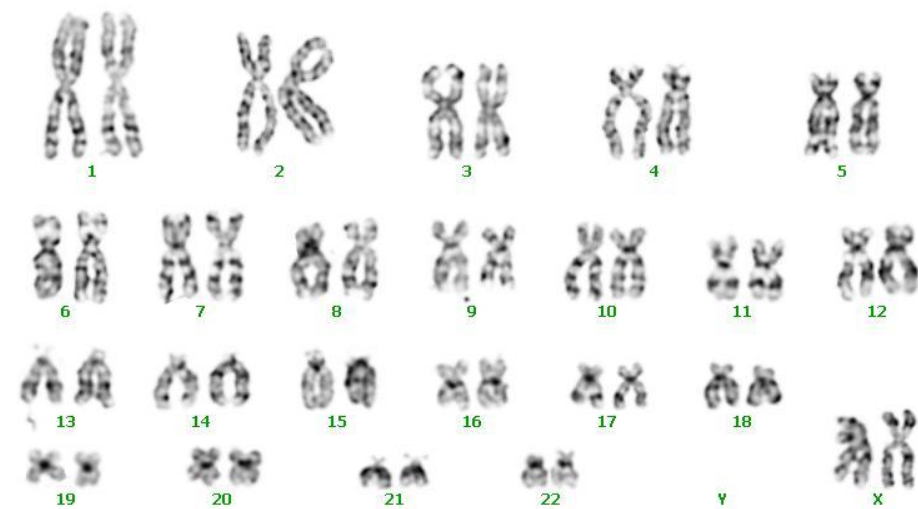
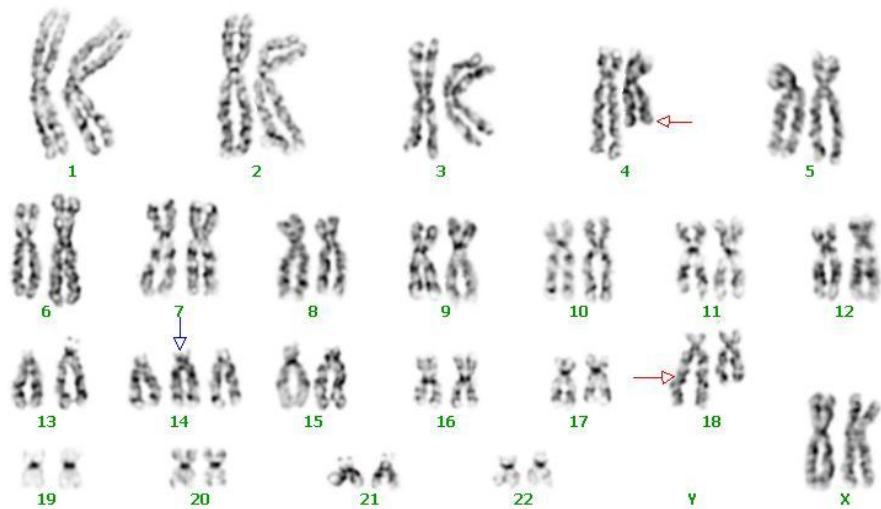
Soft tissue, SCM, right, mass excision:

Fibromyxoid neoplasm, consistent with low grade fibromyxoid sarcoma.

<Note>

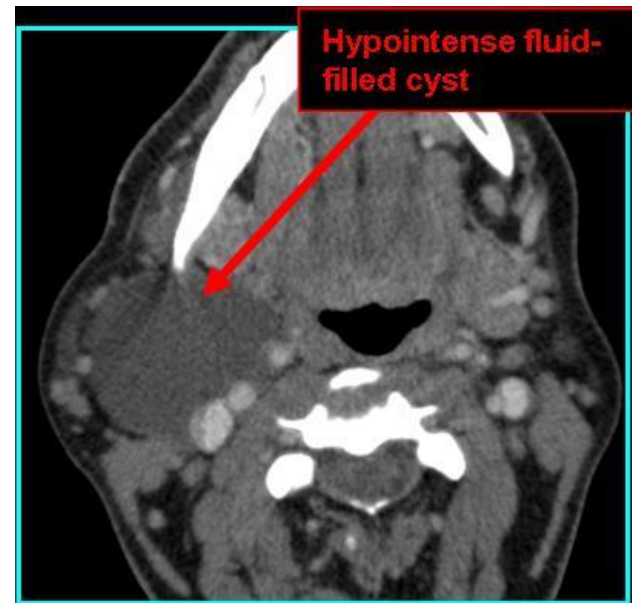
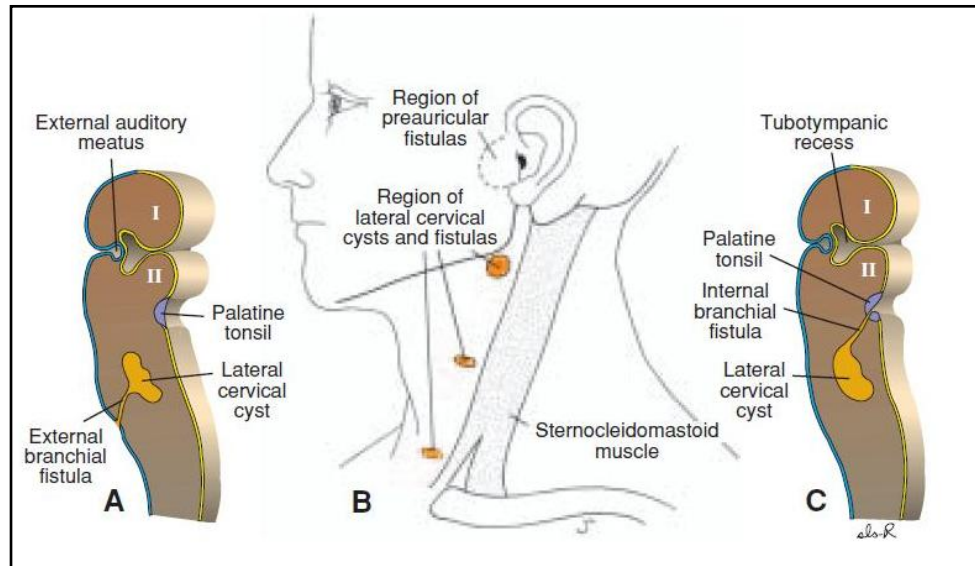
본 종양은 소아에서 생기는 fibromatosis colli, infantile fibromatosis, congenital / infantile fibrosarcoma와의 감별이 필요하나 육안 및 현미경 소견과 immunohistochemistry 소견을 고려할 때 low grade fibromyxoid sarcoma의 가능성이 높을 것으로 생각합니다.

Results of immunohistochemical stain: CD34(negative),  $\beta$ -catenin(inconclusive), Actin(inconclusive), BCL-2(negative), EMA(weak positive), MIB-1(about 7%)



Karyotype of the primary culture cells  
47,XX,t(4;18)(q27;q23),+14[3]/46,XX[27]

# Branchial cleft cyst





# Diagnosis of Congenital Muscular Torticollis

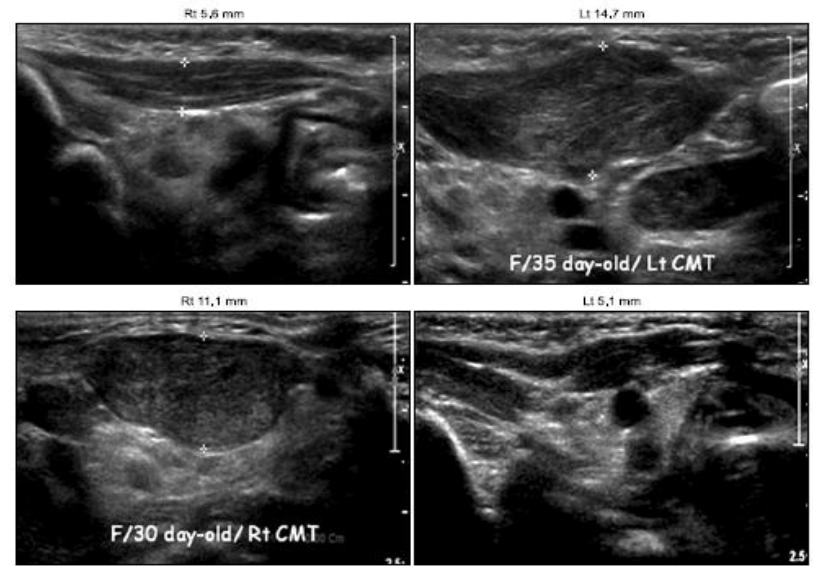
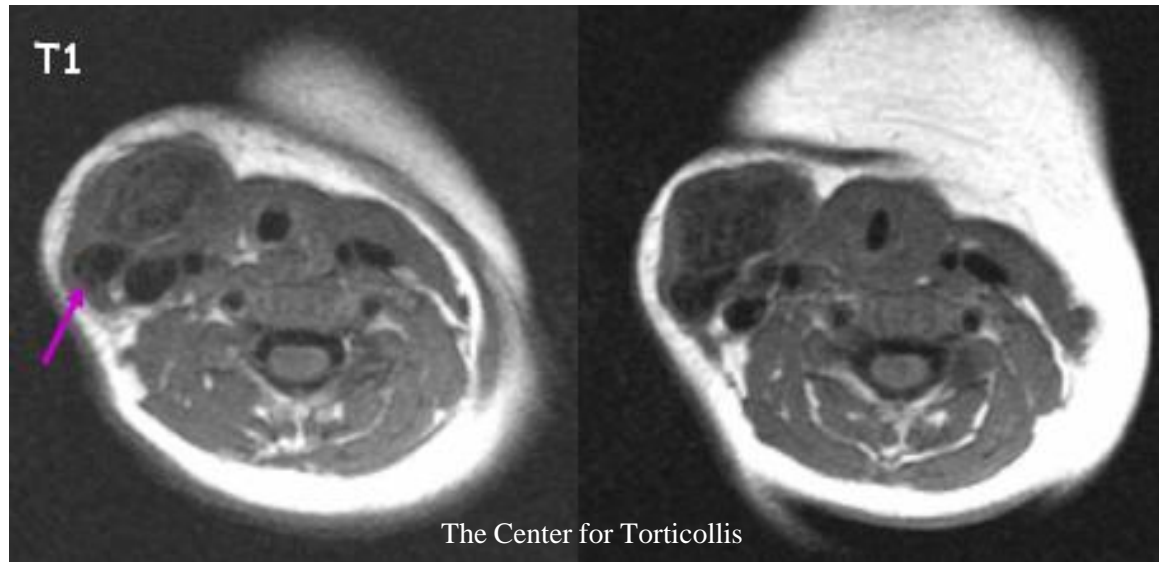


Fig. 1. The ultrasonographic findings of the sternocleidomastoid muscles for the case 1 where sisters had congenital muscular torticollis.



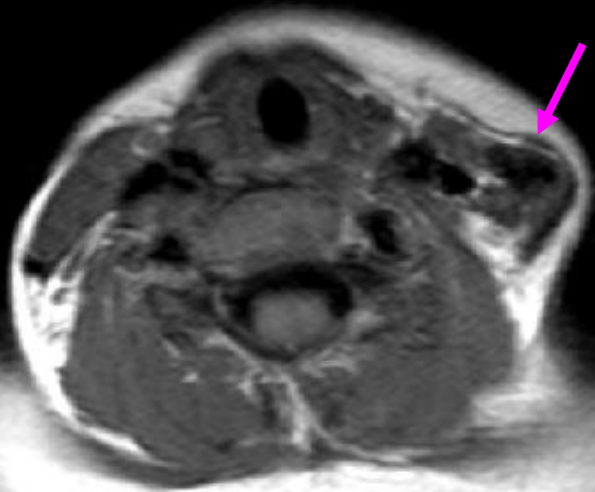
Cho KH, Kim JY, Lee IY, Yim SY. Congenital Muscular Torticollis in Siblings: A case report and literature review. J Korean Acad Rehabil Med. 2009;33:731-734

Yim SY, Lee IY, Park MC, Kim JH. Differential diagnosis and management of abnormal posture of the head and neck. J Korean Med Assoc. 2009; 52: 726

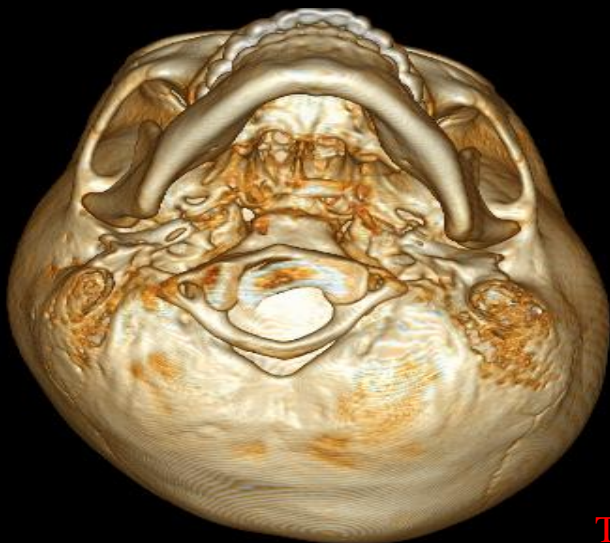




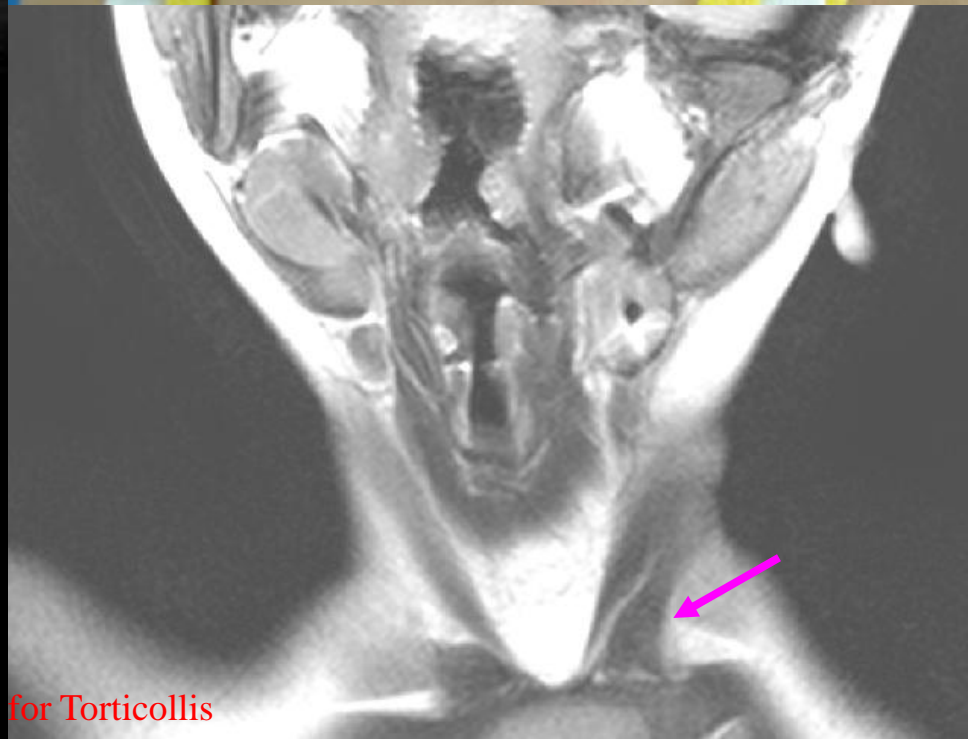
F/ 5.7 year-old  
Left CMT (CH) of Lt. SCM

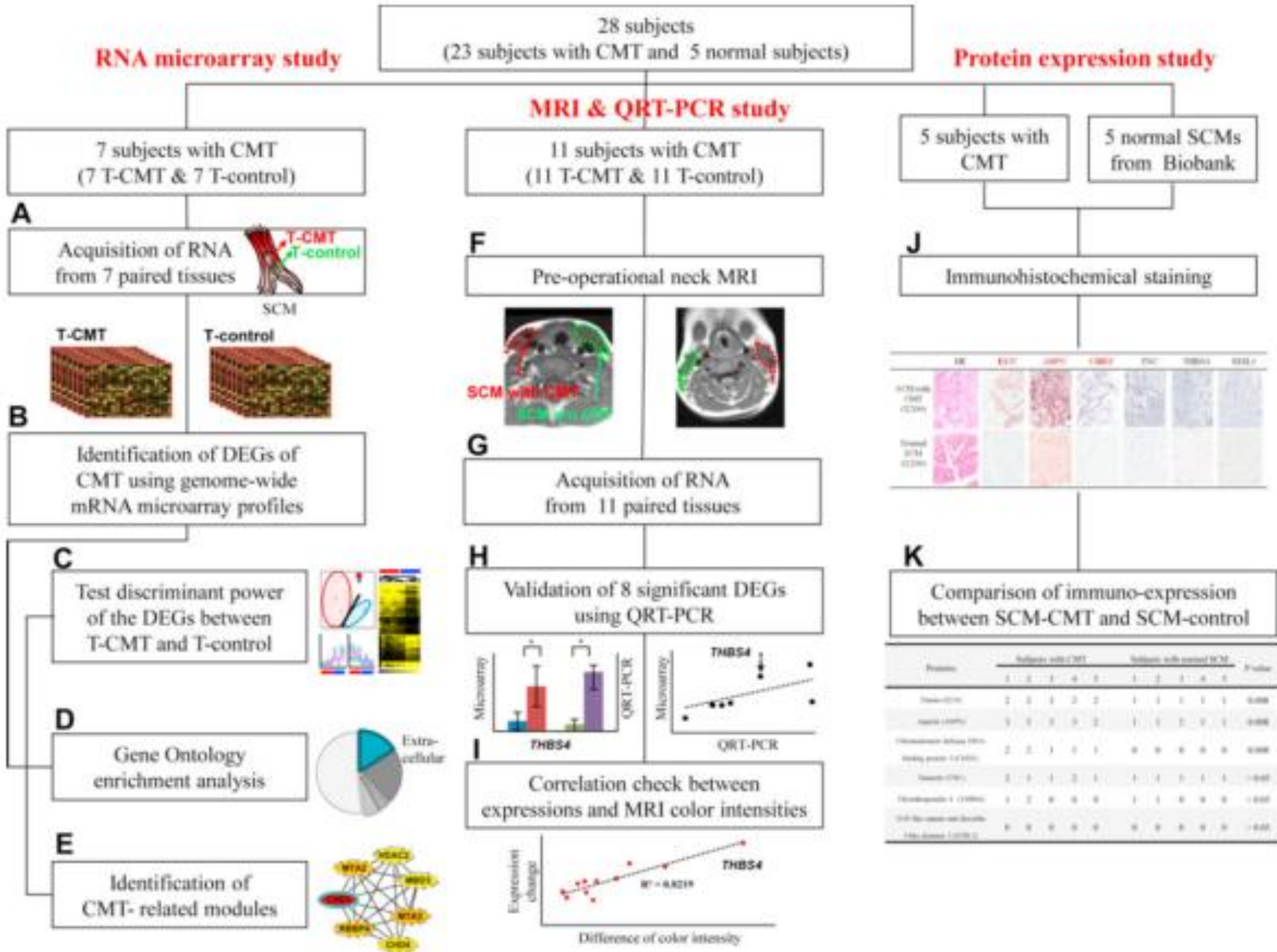


The Center for Torticollis



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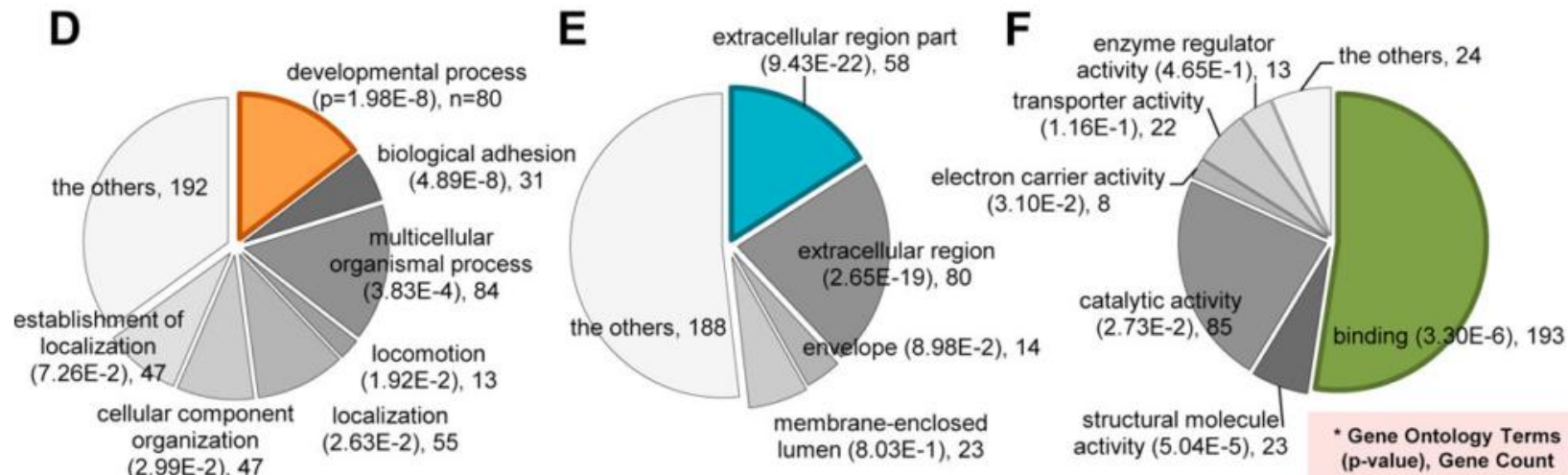


Yim, SY, et al. Integrative analysis of congenital muscular torticollis: from gene expression to clinical significance. BMC Medical Genomics 2013, 6(Suppl 2):S10

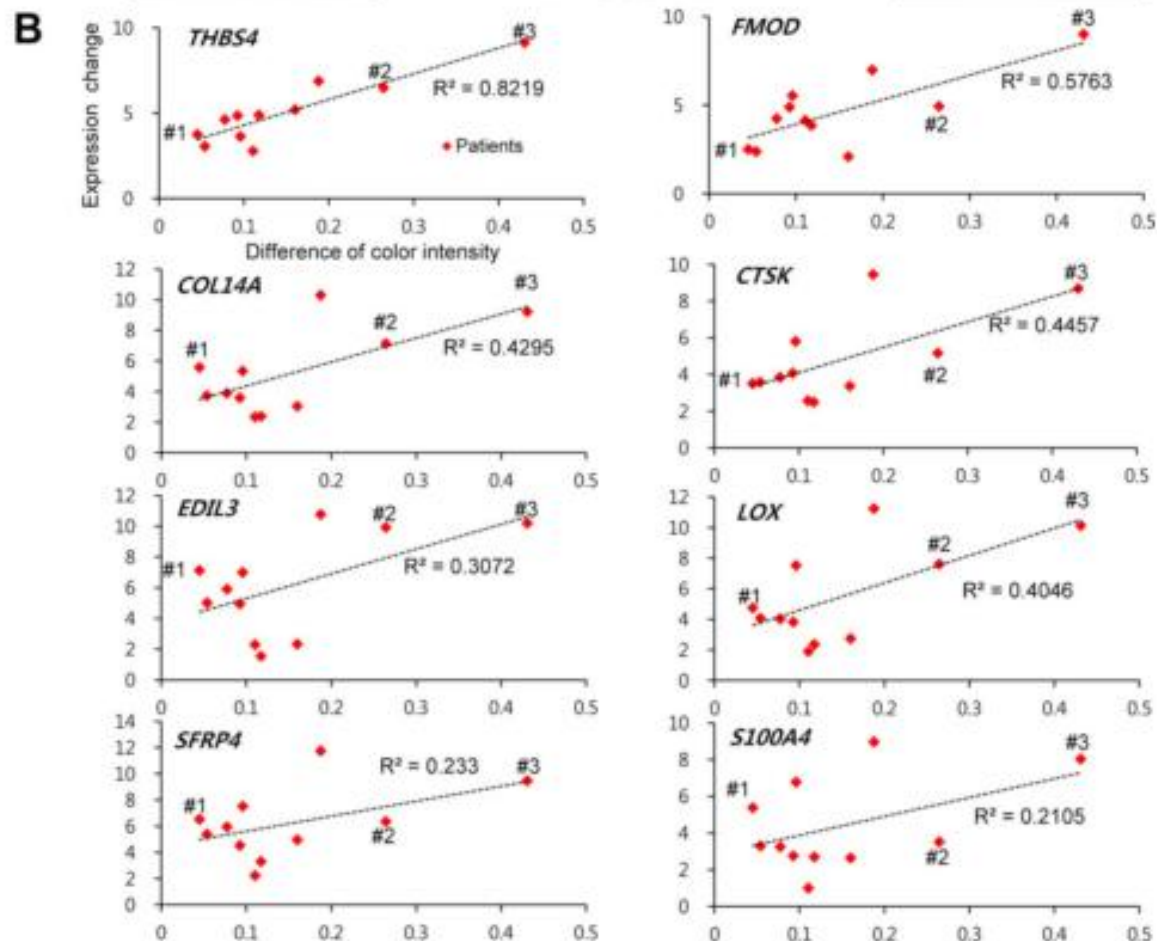
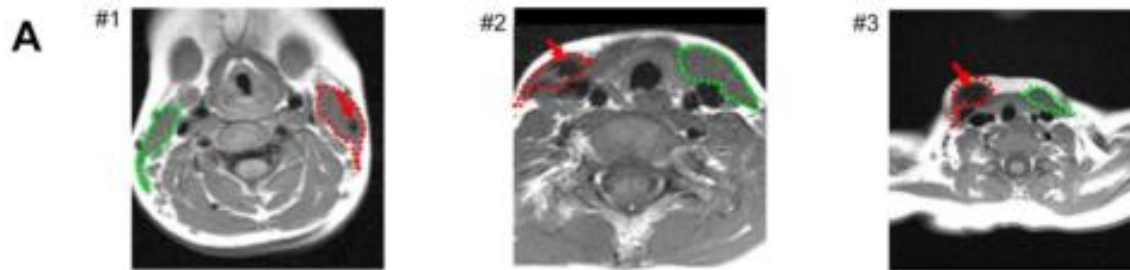
**Table 1 The top-20 over-expressed genes (sorted according to the fold changes).**

Gene symbol	Full name of the gene	Gene ID	Fold change of the expression level	p-value
<b>EDIL3</b>	<b>EGF-like repeats and discoidin I-like domains 3</b>	<b>10085</b>	<b>9.852</b>	<b>0.01</b>
ASPN	asporin	54829	8.240	0.002
<b>THBS4</b>	<b>thrombospondin 4</b>	<b>7060</b>	<b>8.197</b>	<b>0.004</b>
TNMD	tenomodulin	64102	7.576	0.047
NOV	nephroblastoma overexpressed gene	4856	5.583	0.002
SFRP2	secreted frizzled-related protein 2	395546	4.564	0.021
<b>SFRP4</b>	<b>secreted frizzled-related protein 4</b>	<b>6424</b>	<b>4.534</b>	<b>&lt;0.001</b>
MXRA5	matrix-remodelling associated 5	25878	4.506	0.019
<b>FMOD</b>	<b>fibromodulin</b>	<b>2331</b>	<b>4.252</b>	<b>0.014</b>
<b>CTSK</b>	<b>cathepsin K</b>	<b>1513</b>	<b>4.099</b>	<b>0.001</b>
<b>COL14A1</b>	<b>collagen, type XIV, alpha 1</b>	<b>7373</b>	<b>4.022</b>	<b>0.001</b>
<b>LOX</b>	<b>lysyl oxidase</b>	<b>153455</b>	<b>4.010</b>	<b>&lt;0.001</b>
FAM38B	family with sequence similarity 38, member B	63895	4.000	0.016
BGN	biglycan	633	3.764	<0.001
GXYLT2	glucoside xylosyltransferase 2	727936	3.604	0.01
FIBIN	fin bud initiation factor homolog	387758	3.547	<0.001
STEAP2	six transmembrane epithelial antigen of the prostate 2	261729	3.471	0.001
LUM	lumican	4060	3.440	<0.001
DPT	dermatopontin	1805	3.401	0.003
THY1	Thy-1 cell surface antigen	7070	3.390	0.002

Bolds are the DEGs which were double confirmed by QRT-PCR study.

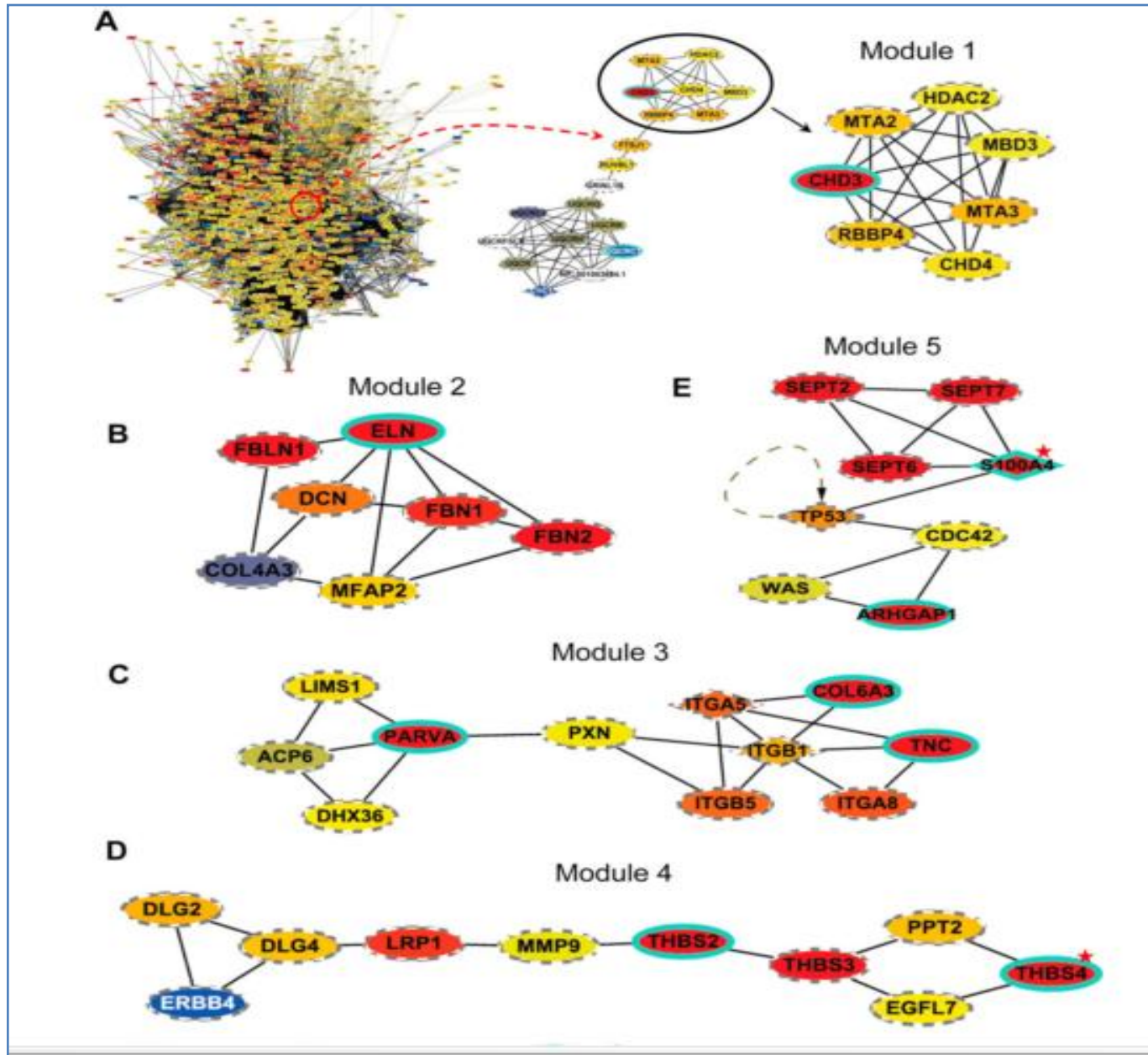






Correlation between the QRT-PCR findings and the MRI findings

# Five CMT-related network modules





## Definition of CMT

The gene expression signatures of CMT is characterized by the over-expression of collagen and elastin fibrillogenesis along with the evidence of DNA repair mechanism and the cytoskeletal rearrangement possibly related with mechanical damage.

Based on the results of this study, CMT might be defined as a developmental disorder of the SCM characterized by fibrosis, ending up with the shortening of the SCM.

The Center for Torticollis

Yim, SY, et al. Integrative analysis of congenital muscular torticollis: from gene expression to clinical significance. BMC Medical Genomics 2013, 6(Suppl 2):S10

# Pathogenesis of CMT



아주대학교병원 재활의학과  
사경센터 · 아동재활클리닉

Physical Medicine & Rehabilitation, Ajou University Hospital  
The Center for Torticollis · The Children's Rehabilitation Clinic

Venous occlusion resulting ischemia in SCM 허혈  
Sequela of compartment syndrome 구획증후군  
Hematoma 혈종...



Congenital muscular torticollis  
근성사경

The Center for Torticollis

## Injury of Skeletal Muscle

1. Disease such as muscular dystrophy
2. Exposure to myotoxic agents, such as bupivacaine or lidocaine
3. Sharp or blunt trauma, such as punctures or contusions
4. Ischemia
5. Exposure to hot or cold temperatures
6. The muscle's own contraction
7. Excessive stretching

Karalaki M, Fili S, Philippou A, Koutsilieris M.

Muscle regeneration: cellular and molecular events. In Vivo 2009;23:779-96

# Concurrent conditions with CMT

Unilateral developmental dysplasia of the hip 12%: intrauterine malposition

Brachial plexus injury up to 5.4%???

Clavicular fracture 2%

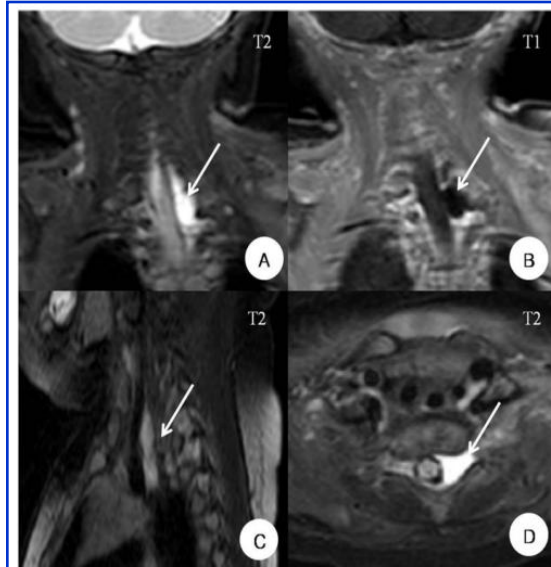


Fig. 1. Brachial plexus MRI. Coronal view of T2-weighted images (A) and T1-weighted images (B), sagittal view of T2-weighted images (C), and axial view of T2-weighted (D) images. High signal intensities on (A), (C), (D) and low signal intensity on (B) show pseudocysts (arrows) at the left sides of the C6, C7, and T1 levels that resulted from tearing of the neural sheath at the age of 1 month.

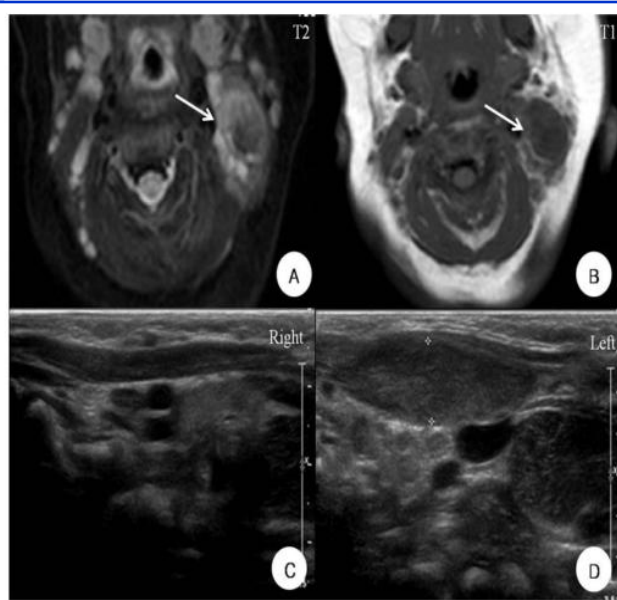
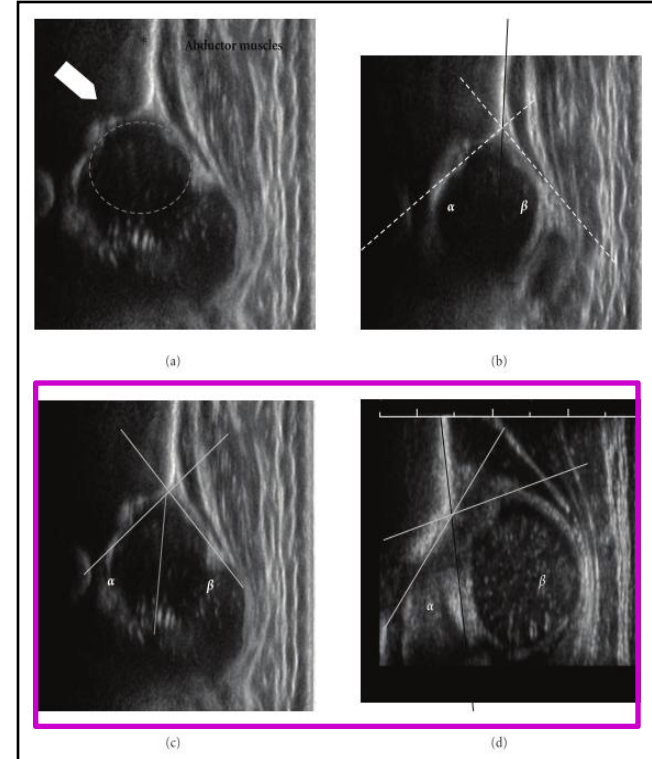


Fig. 2. Neck MRI showing left congenital muscular torticollis (arrows at A, B) with high signal intensity on an axial T2-weighted image (A) and isosignal intensity on an axial T1-weighted image (B). Ultrasonographic images (C, D) show the thickened left sternocleidomastoid muscle.



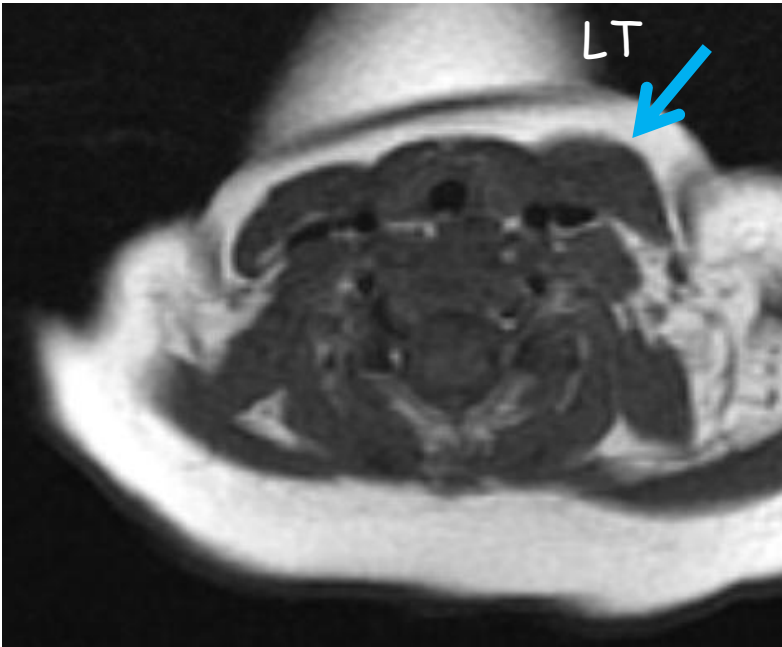
## The Center for Torticollis

Lee HB, Park MC, Kim C, et al. Concurrence of Obstetric Brachial Plexus Injury, Congenital Muscular Torticollis and Cleft Palate.

J Genet Med 2011; 8: 71-75.



**Fig. 1.** Concurrence of right clavicular fracture and left congenital muscular torticollis.



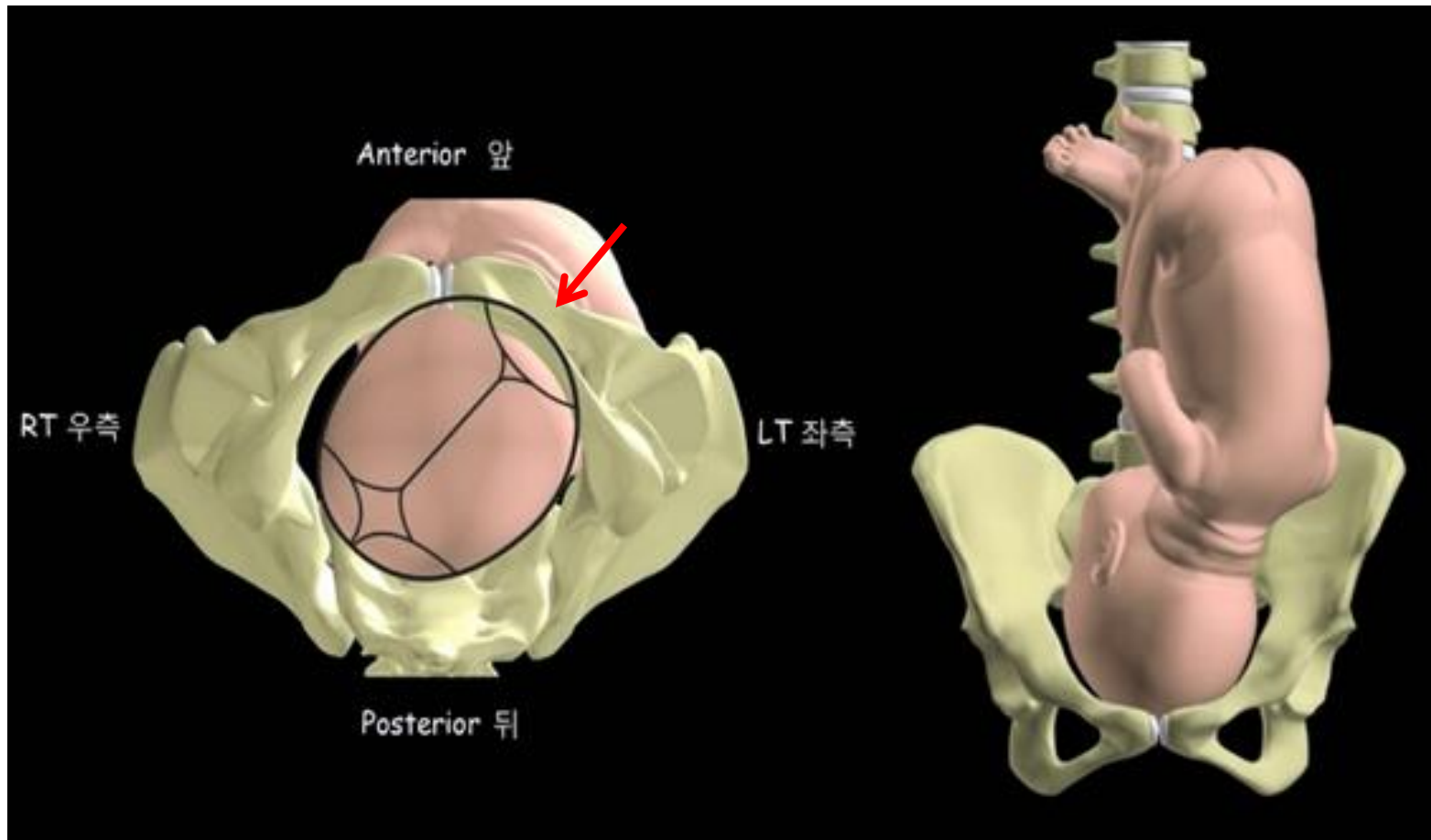
In our clinical series of 996 subjects with CMT, concurrence of CMT and clavicular fracture was found in 20 subjects (2.01%), where CMT and clavicular fracture occurred on the contralateral side for each other in 18 out of 20 (90%) rather than the same side.

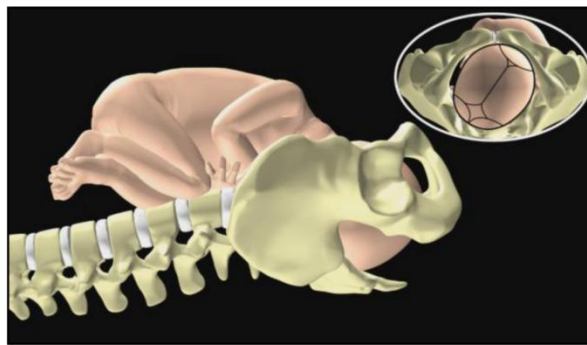
Contingency table between the location of congenital muscular torticollis and the location of clavicular fracture.

Location of congenital muscular torticollis	Location of clavicular fracture	
	Right	Left
Right	1	9
Left	9	1

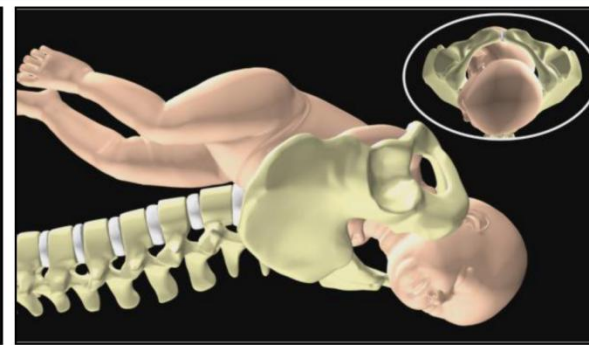
The Center for Torticollis/ unpublished data

**Left occipito-anterior position or the LOA position**  
fetal occiput in left anterior position of maternal pelvis  
the most common and the best fetal position for delivery

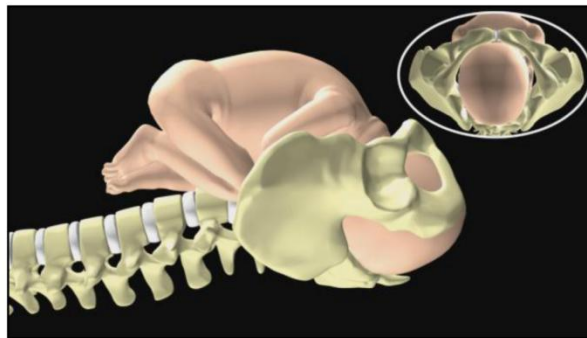




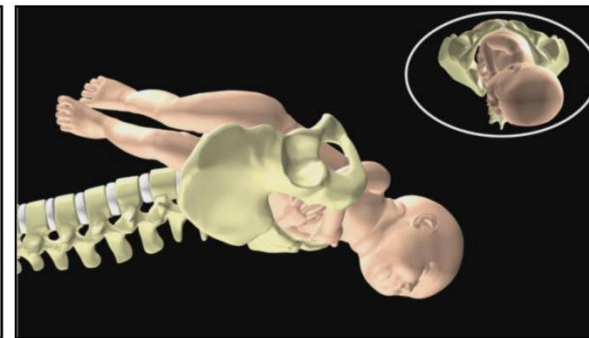
1. Left occipito-anterior fetal position



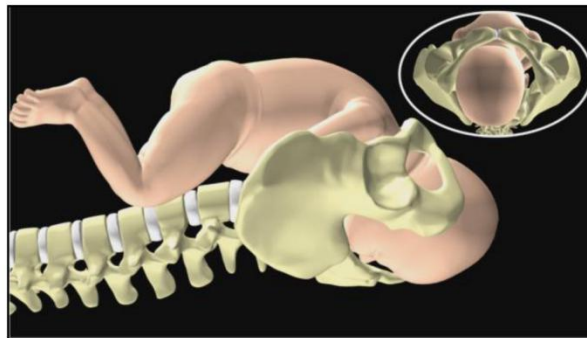
4. External rotation of the shoulders



2. Simultaneous internal rotation of both head and shoulders

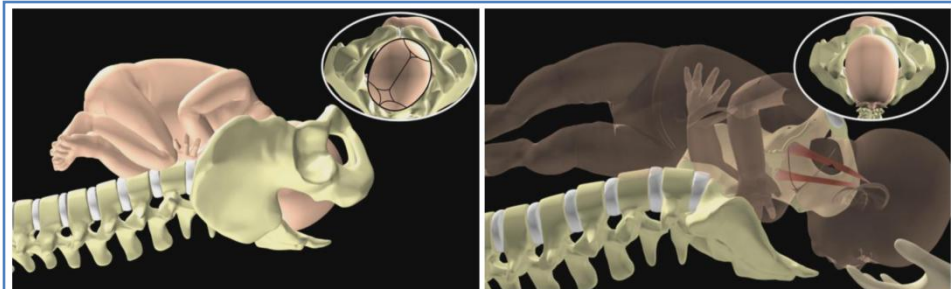


5. Delivery of the shoulders



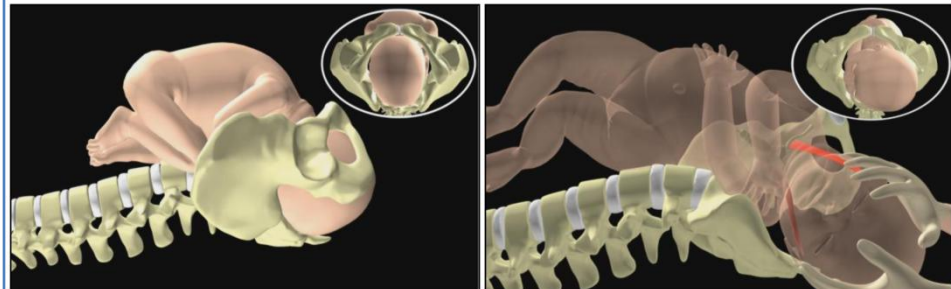
3. Delivery of the head





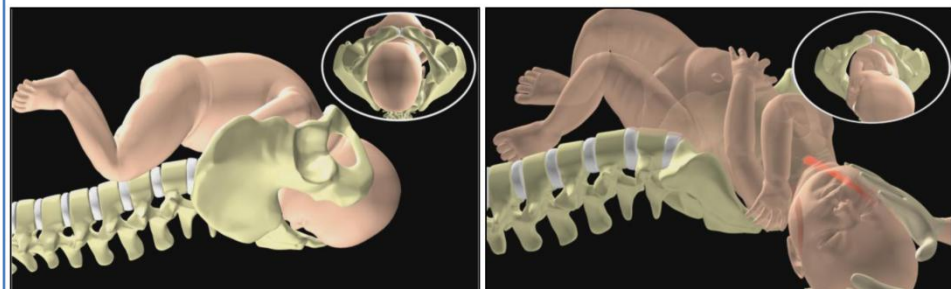
1. Left occipito-anterior fetal position

4. Delayed shoulder expulsion



2. Simultaneous internal rotation of both head and shoulders

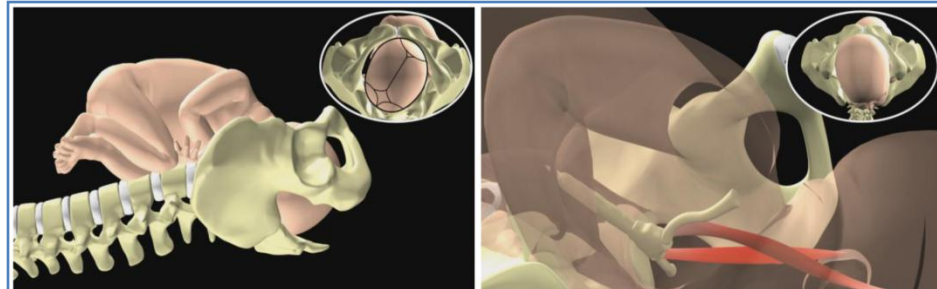
5. Downward traction of the baby



3. Delivery of the head

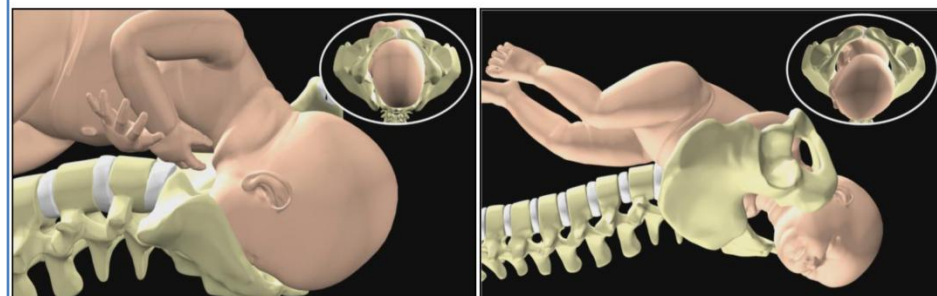
6. Delivery of the shoulders with RCMT

A proposed pathogenesis of concurrence of right congenital muscular torticollis (RCMT) during vaginal delivery in left occipito-anterior position.



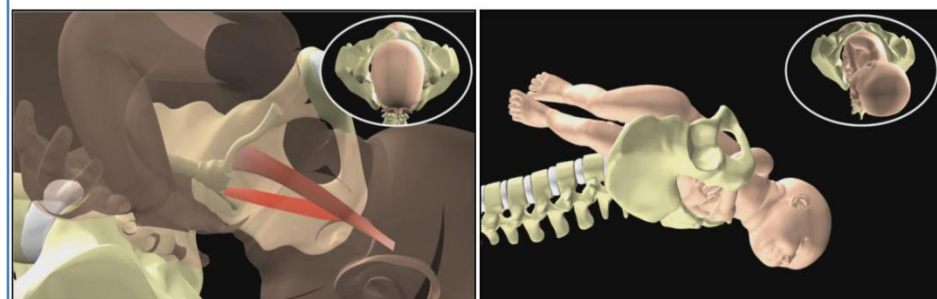
1. Left occipito-anterior fetal position

4. Delivery of the head with LCMT



2. Solitary internal rotation of the head

5. External rotation of the shoulders



3 Overstretching of LSCM

6. Delivery of the shoulders

A proposed pathogenesis of concurrence of left congenital muscular torticollis (LCMT) during vaginal delivery in left occipito-anterior position. LSCM, left sternocleidomastoid muscle.



Prenatal period 분만전기

Perinatal period 주산기



Excessive stretching of unilateral SCM

편측 흉쇄유돌근 과도한 신전

Venous occlusion resulting ischemia in SCM 허혈

Sequela of compartment syndrome 구획증후군

Hematoma 혈종...

Congenital muscular torticollis

근성사경

**Table 1.** Characteristics of the Children with CMT and Who Were younger than 6 Years-old Based on the Method of Child Birth\*

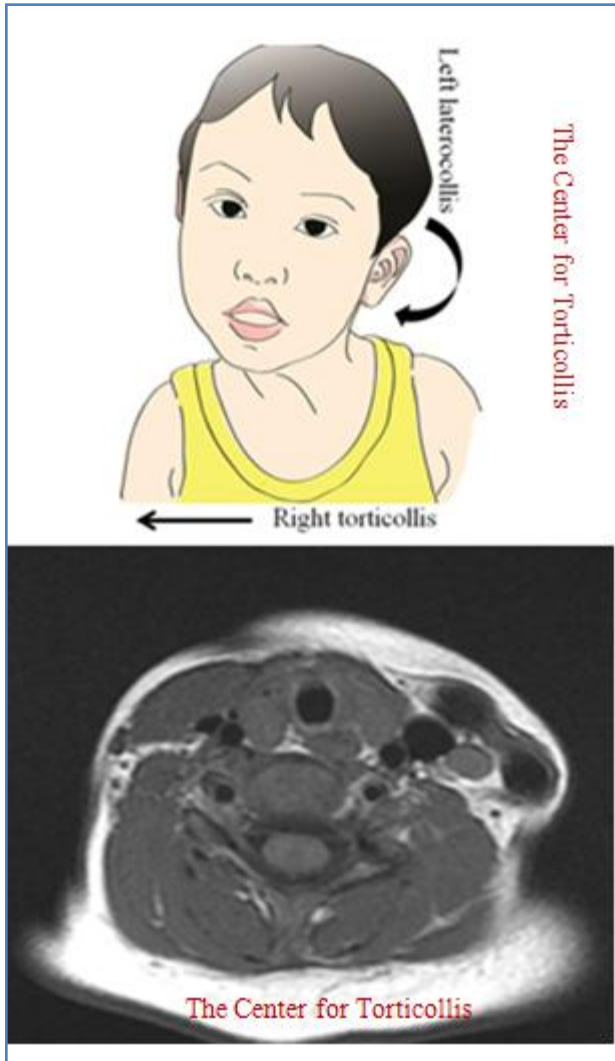
Characteristics	Vaginal delivery	Cesarean section	p-value
Number of subjects (%)	132 (74.16)	46 (25.84)	0.000
Age at the first visit (months-old) <sup>†</sup>	5.84±9.15	5.45±9.59	0.824
Gestational age (weeks) <sup>†</sup>	38.97±2.90	38.79±1.12	0.335
Birth weight (gram) <sup>†</sup>	3,302±385	3,173±439	0.292
Percentage of subjects who needed stretching exercises for CMT (number of subjects)	60.77 (79)	64.58 (31)	0.729
Percentage of subjects who needed surgical release for CMT (number of subjects)	20.76 (27)	20.83 (10)	1.000

\*<6-years-of-age, n=178. <sup>†</sup>mean±standard deviation

Lee SJ, Han JD, Lee HB, Hwang JH, Kim SY, Park MC, Yim SY. Comparison of clinical severity of congenital muscular torticollis based on the method of child birth.

Ann Rehabil Med 2011;35:641-7

# Symptoms and signs of congenital muscular torticollis



Kim JK, Yim SY. Clinical characteristics of abnormal postures of the head and neck caused by unilateral superior oblique palsy. J Korean Acad Rehabil Med 2011; 35: 272-278



The Center for Torticollis

The maneuver inducing the cough reflex

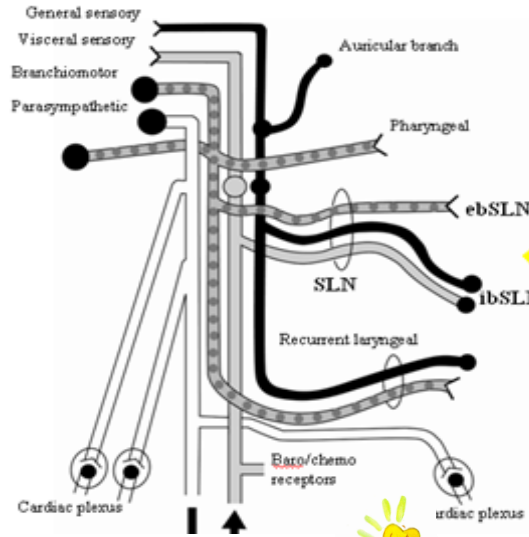


Yim SY, Lee IY, Cho KH, Kim JK, Lee IJ, Park MC: The laryngeal cough reflex in congenital muscular torticollis: Is it a new finding?

Am J Phys Med Rehabil 2010;89:147–152.



# Laryngeal cough reflex

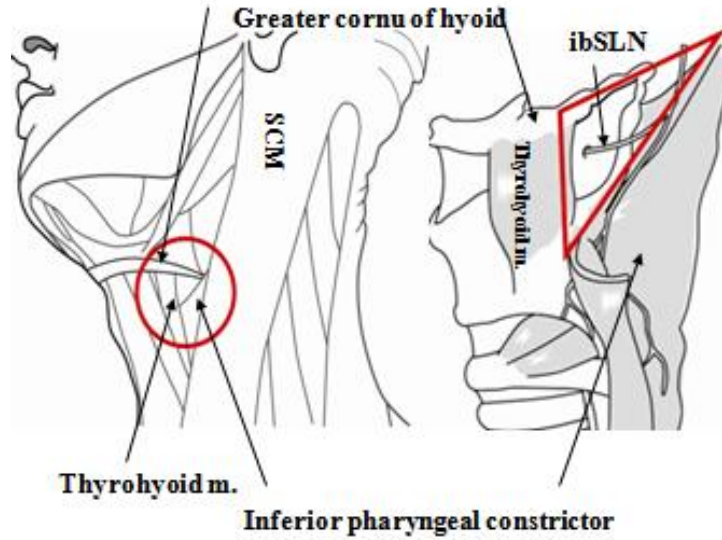


Brainstem cough centers  
in the vicinity of the  
nucleus tractus solitarius

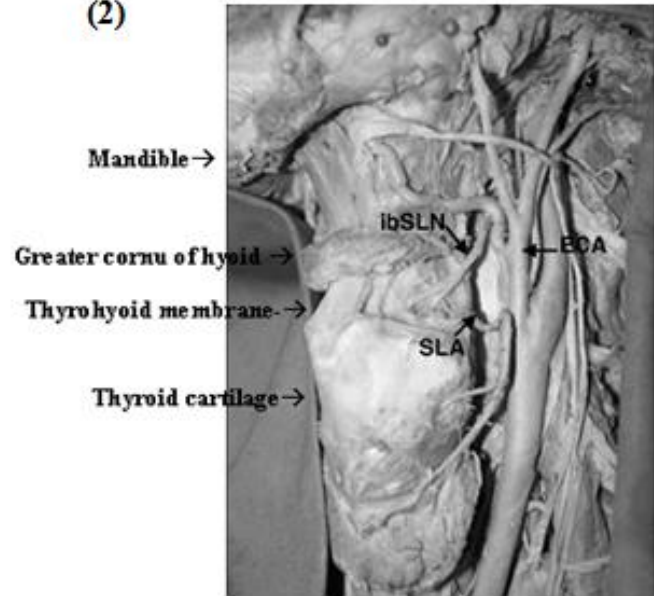


Efferent fibers in  
vagus, phrenic &  
intercostal n

(1)



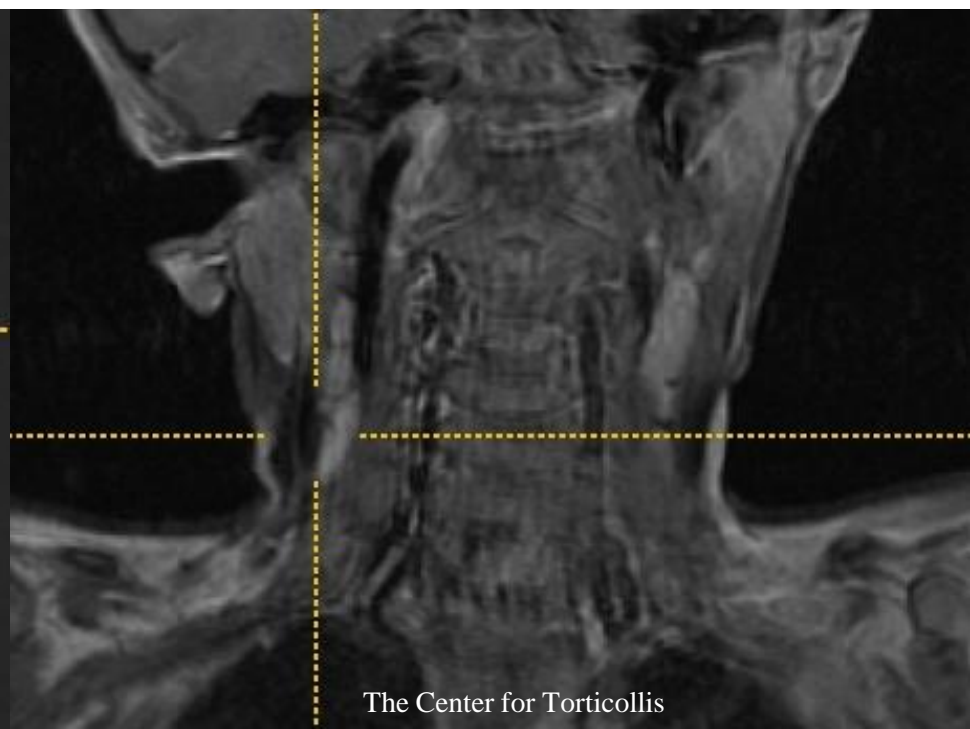
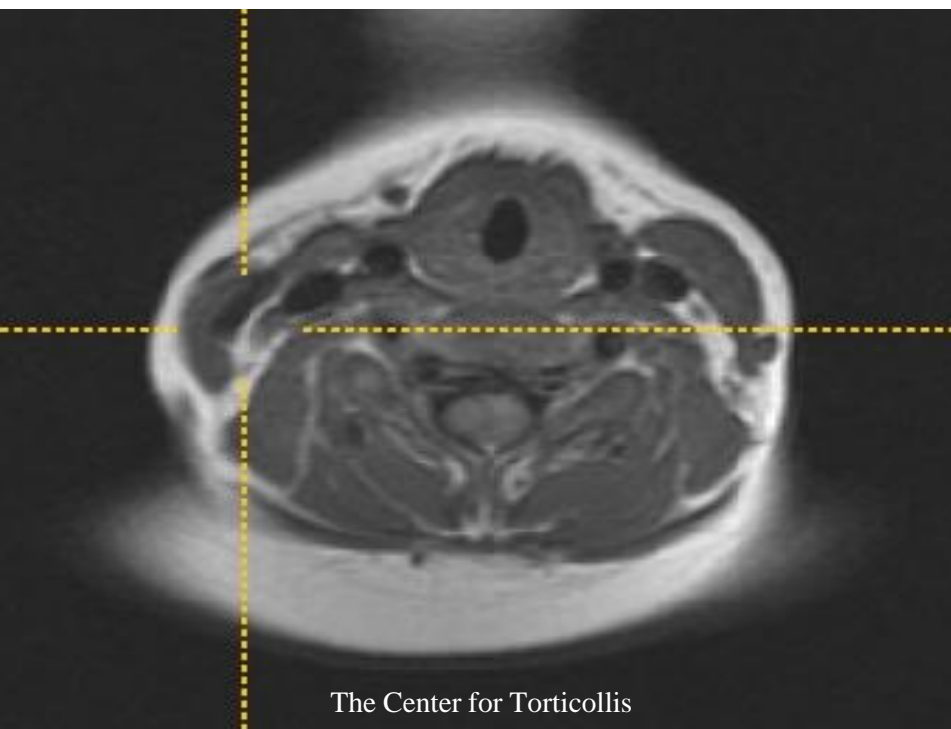
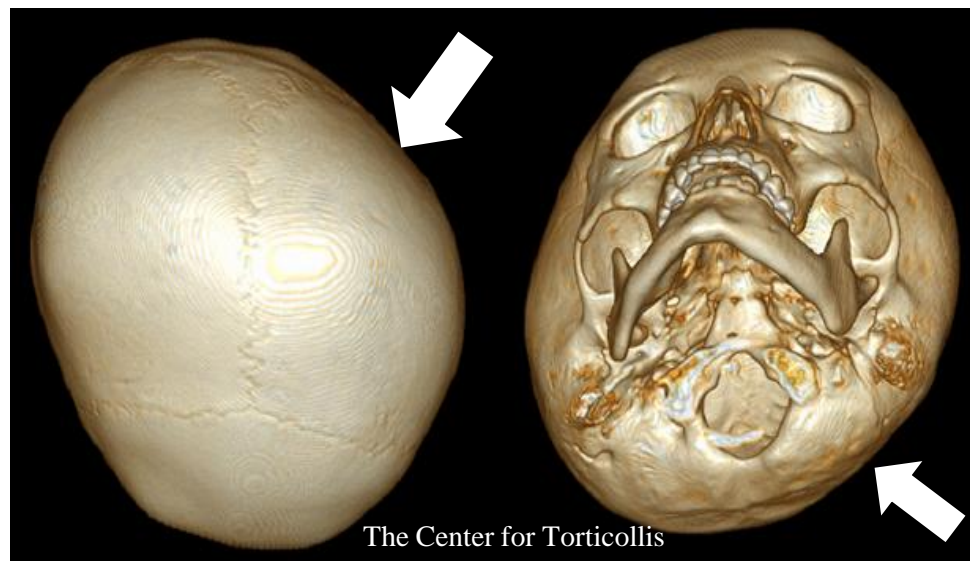
(2)



Yim SY, Lee IY, Cho KH, Kim JK, Lee IJ, Park MC: The laryngeal cough reflex in congenital muscular torticollis: Is it a new finding? Am J Phys Med Rehabil 2010;89:147-152.



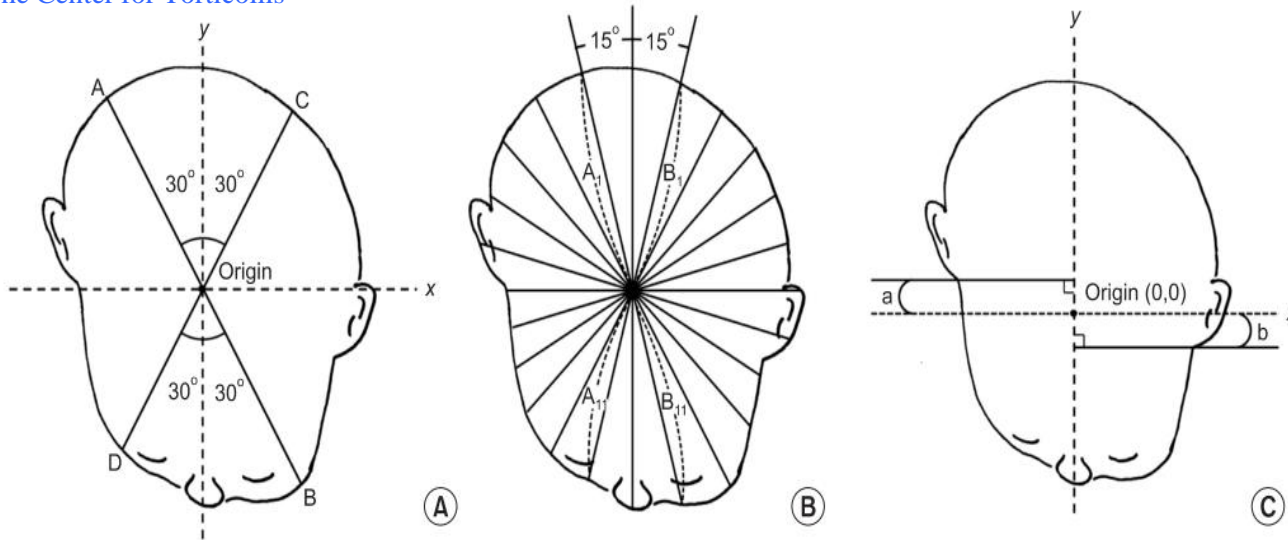
F/3.8 yr  
Right CMT (S+ C)  
Left plagiocephaly





## Ear shift and acquired ear deformity associated with CMT

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**Fig. 3.** (A) Diagonal difference (DD; mm)=longer diagonal (AB)-shorter diagonal (CD). Cranial vault asymmetry index (%)=DD/CD×100. (B) Radial symmetry index (mm)=|(A<sub>1</sub>+A<sub>2</sub>+...+A<sub>11</sub>)-(B<sub>1</sub>+B<sub>2</sub>+...+B<sub>11</sub>)|. (C) The ear shift (mm)=|a-b|.



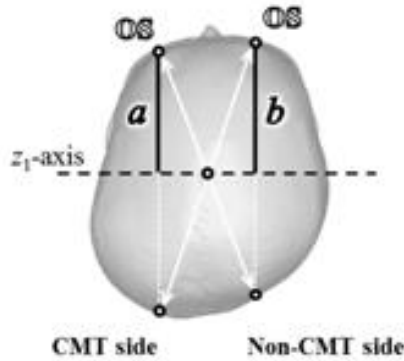
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Kim SY, Park MS, Yang, JI , Yim SY.

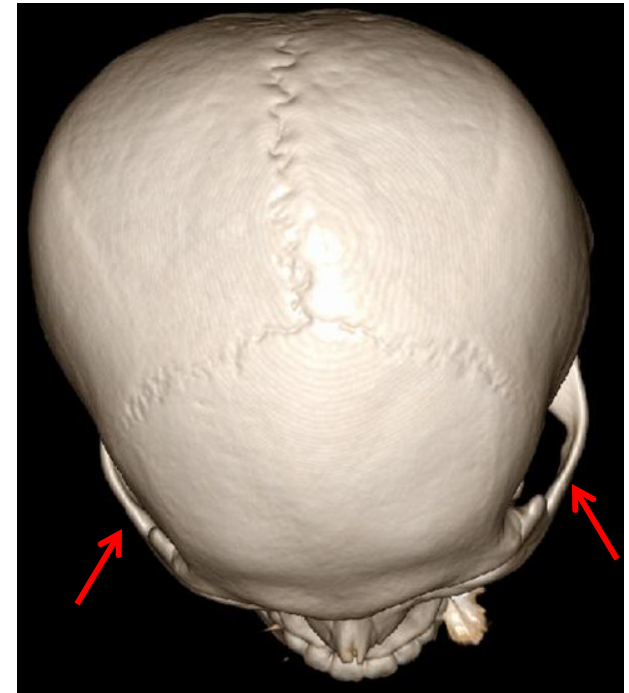
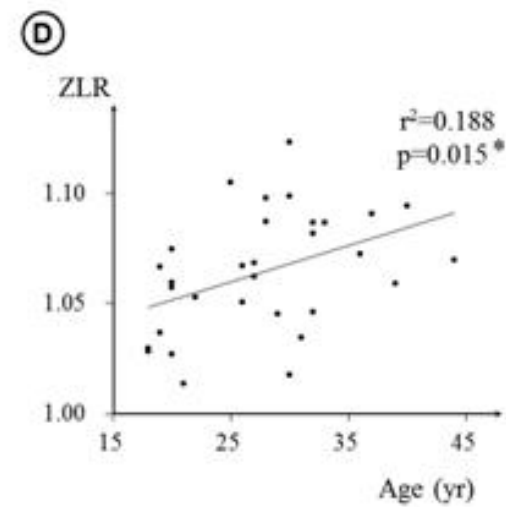
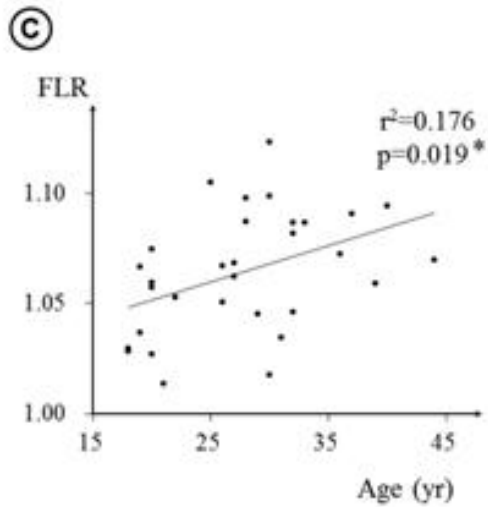
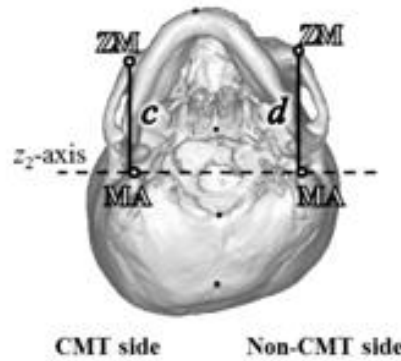
Comparison of helmet therapy and counter positioning for deformational plagiocephaly.

Ann Rehabil Med 2013; . **37: 785-795**

Ⓐ Frontal length ratio (FLR)  
=  $b/a$



Ⓑ Zygomatic length ratio (ZLR)  
=  $d/c$



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Measurement of the anteroposterior length asymmetry on the axial plane.

# Musculoskeletal complications of CMT

1. Ipsilateral laterocollis
2. Contralateral torticollis
3. Contralateral deformational plagiocephaly

Ipsilateral flattening of forehead and malar bone

Posterior displacement of ipsilateral ear

4. Elevation of ipsilateral shoulder

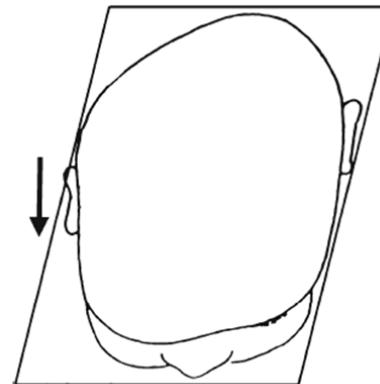
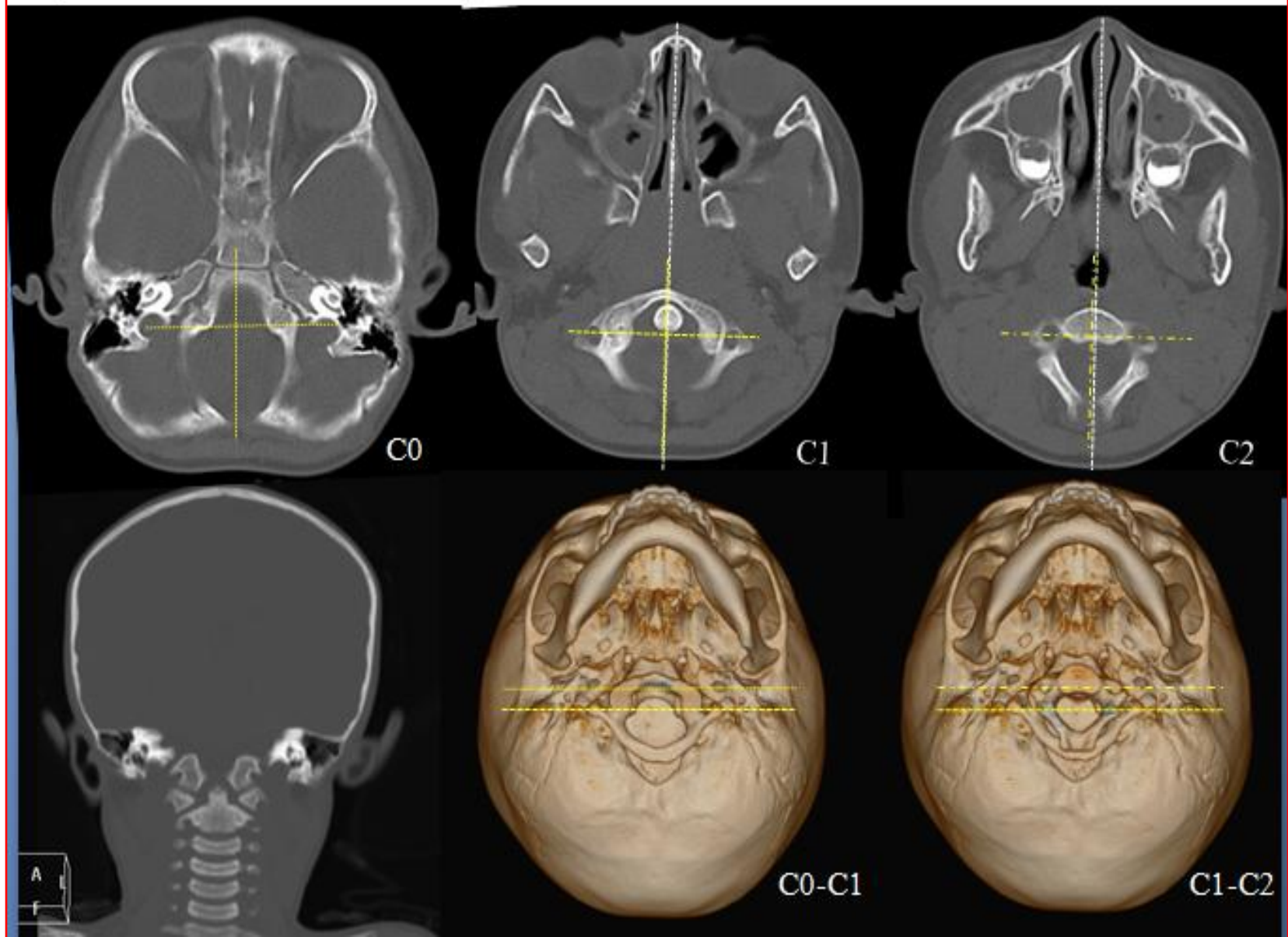


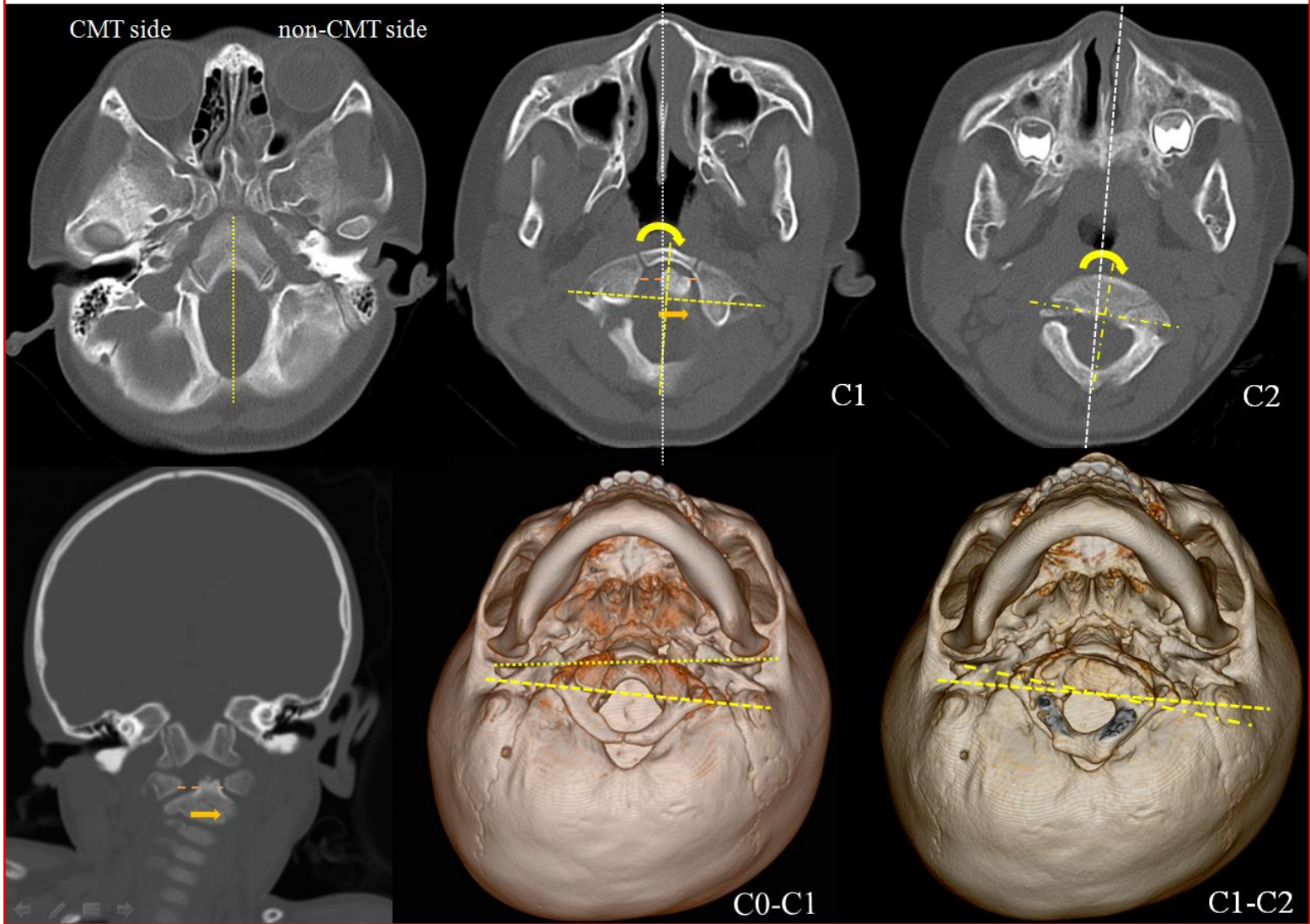
Figure 3. 48 months-old male without congenital muscular torticollis



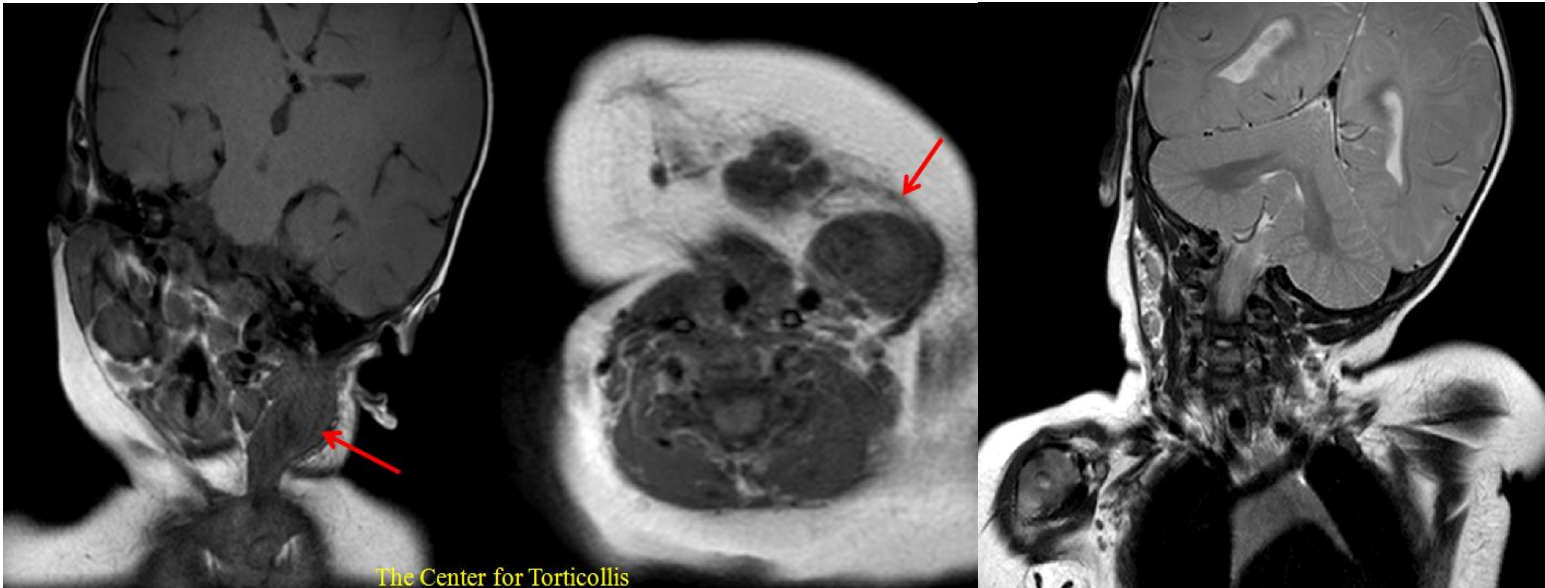
The Center for Torticollis/ unpublished data



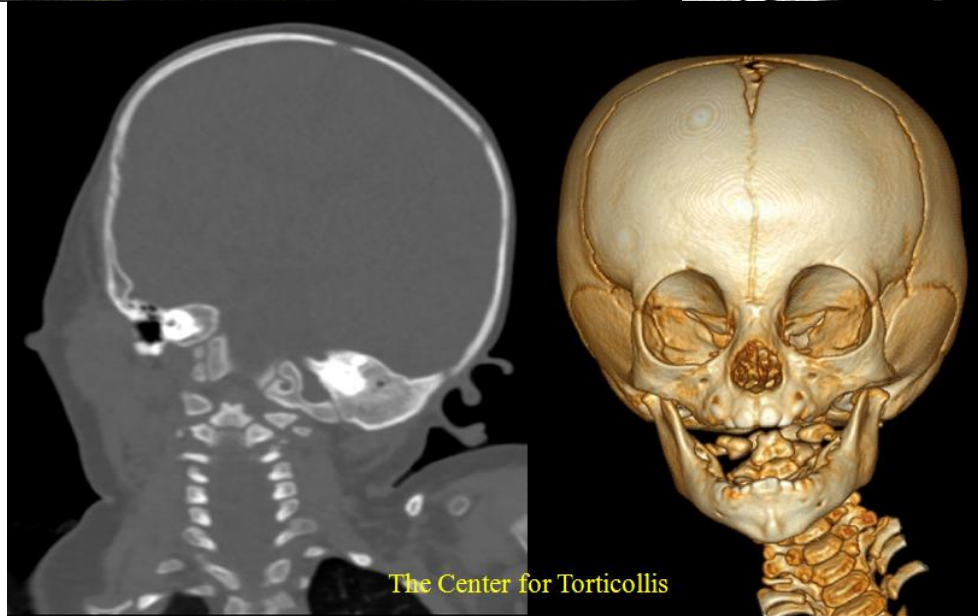
Figure 2. 46 months-old male subject with right congenital muscular torticollis with multiple craniovertebral junction abnormalities







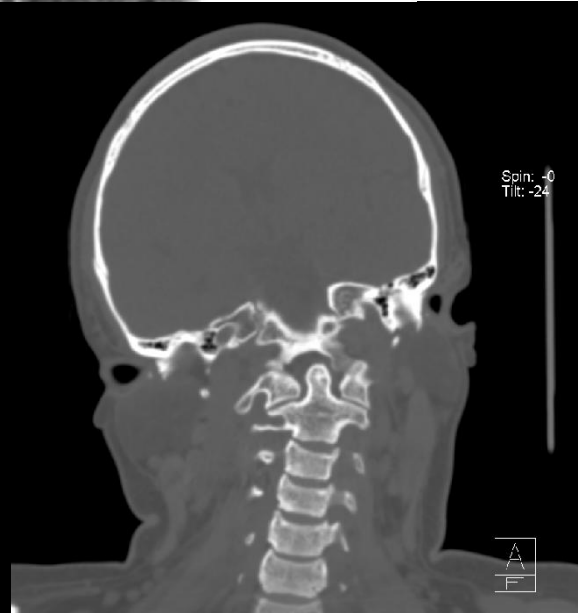
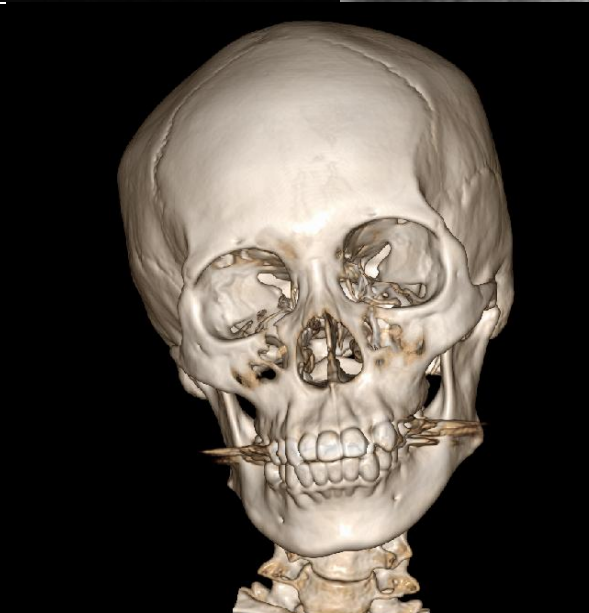
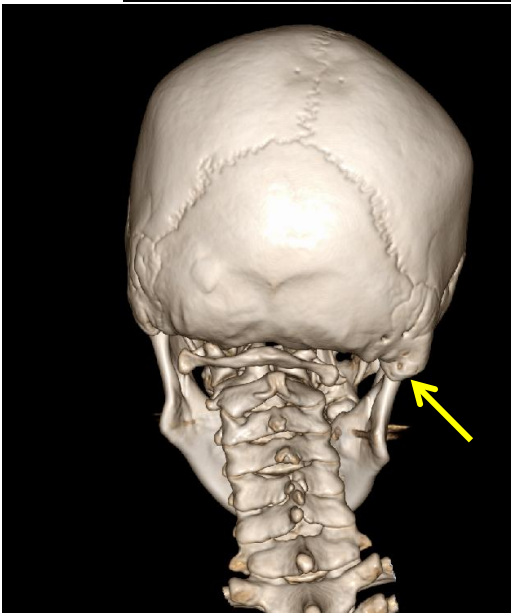
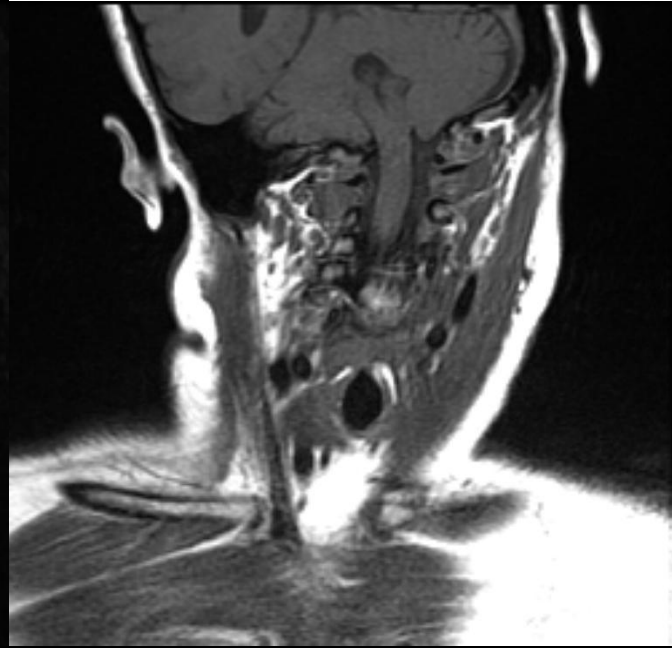
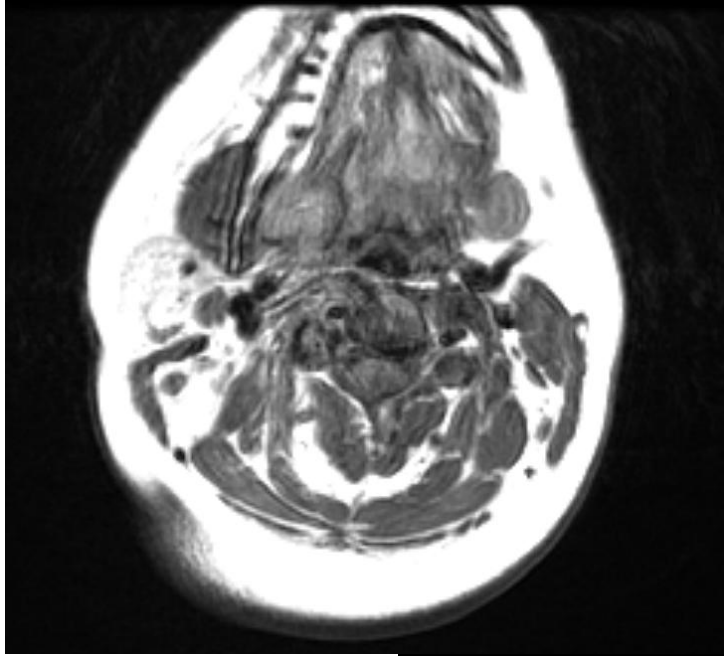
The Center for Torticollis

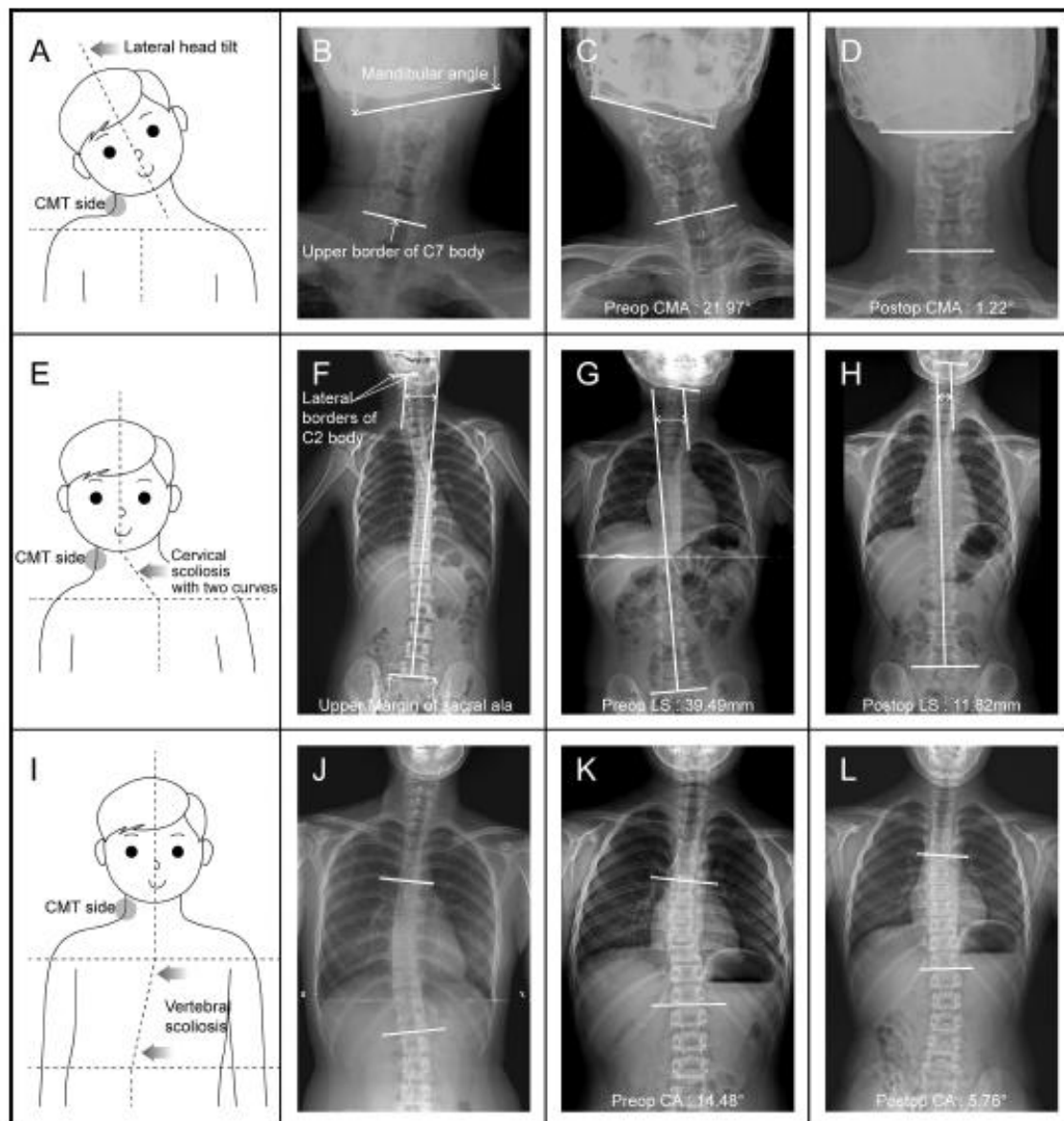


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F/9mo  
LCMT with myelopathy

F/ 24 year-old/ RCMT





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Effectiveness of surgical release in patients with neglected congenital muscular torticollis according to age at the time of surgery

Ann Rehabil Med (in press)

# Management of Congenital Muscular Torticollis



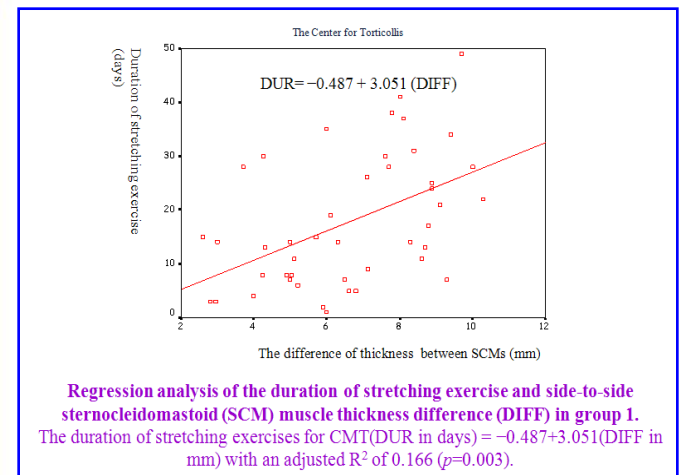
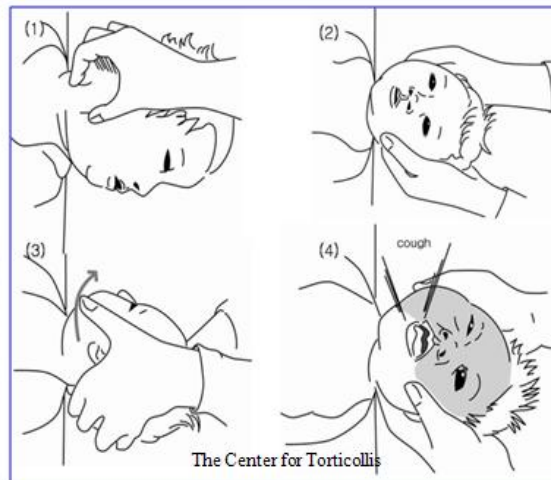
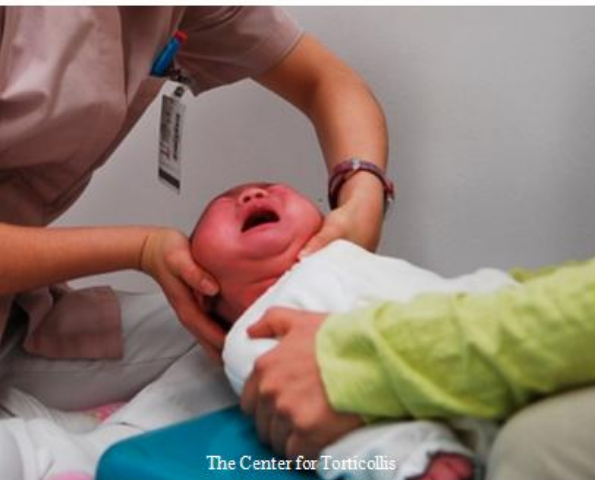
아주대학교병원 재활의학과  
사경센터 · 아동재활클리닉

Physical Medicine & Rehabilitation, Aju University Hospital  
The Center for Torticollis · The Children's Rehabilitation Clinic

# Manual stretching exercise, massage, ultrasound

## The sooner, the better!

## Restoration of length of SCM



Han JD, Kim SH, Park MC, Yim SY. Thickness of sternocleidomastoid muscle as a prognostic factor of congenital muscular torticollis. *Ann Rehabil Med* 2011; 35: 361-368.

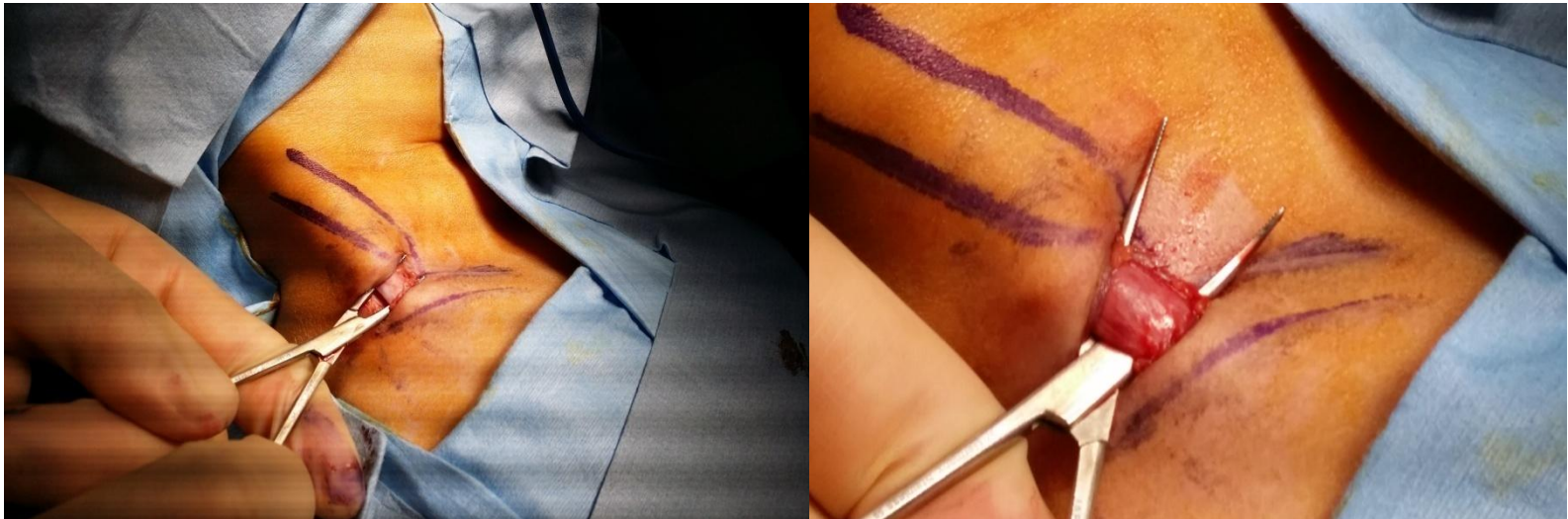
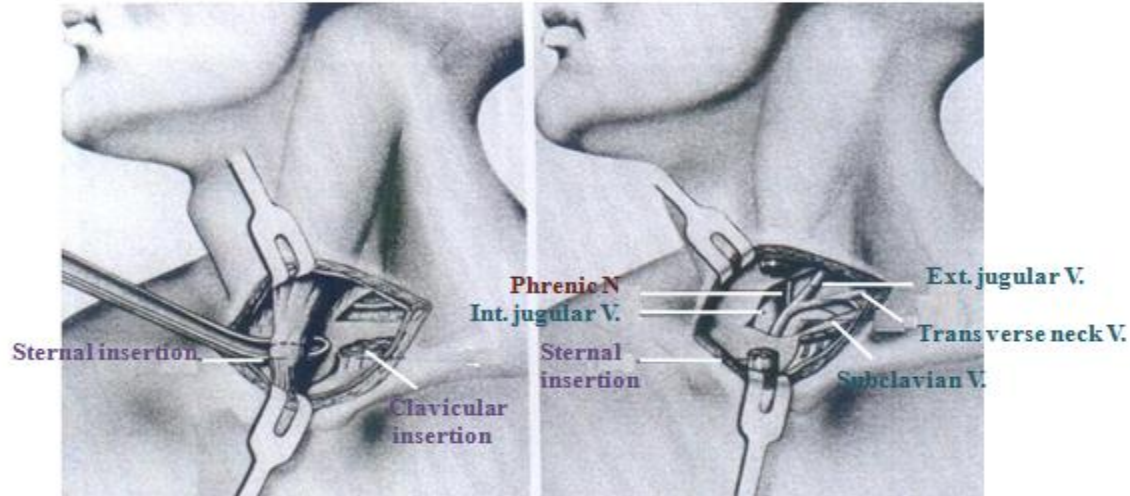
Yim SY, Lee IY, Cho KH, Kim JK, Lee IJ, Park MC: The laryngeal cough reflex in congenital muscular torticollis: Is it a new finding? *Am J Phys Med Rehabil* 2010;89:147-152.



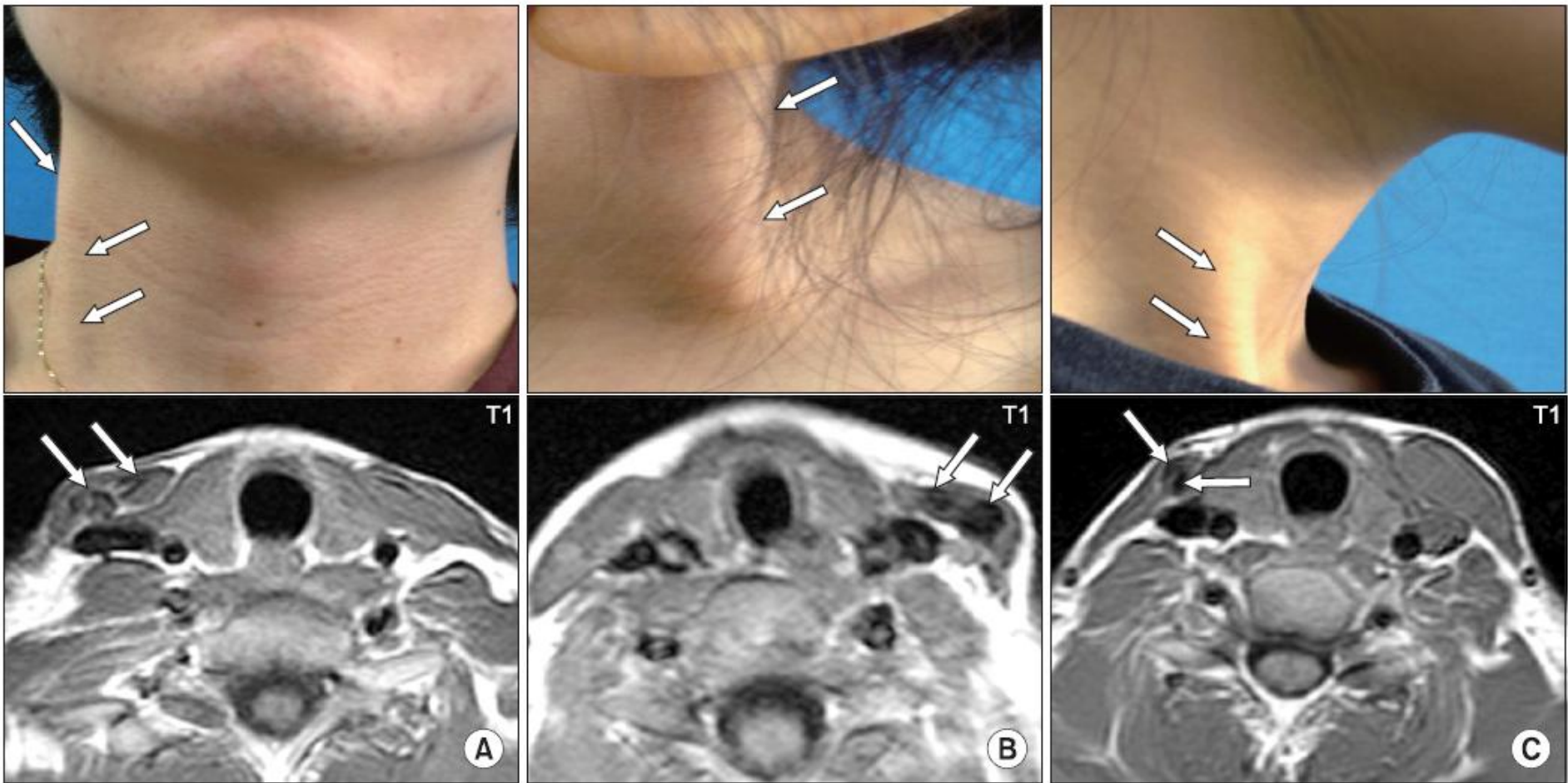
# Unipolar release at the clavicular and sternal insertion: Most commonly used method



J Korean Soc Plast Reconstr Surg 2009; 35: 38- 45



With courtesy of Dr. Jae H. Cho ,  
Dept. of Orthopedics, Ajou Univ. Medical Center



**Fig. 5.** Pictures showing subjects with atrophy or same thickness of the shortened sternocleidomastoid muscle (SCM) with low signal intensities on the T1-weighted axial images of the SCM. (A) Twenty-two year old man with right congenital muscular torticollis (CMT) showing a cord-like right SCM (arrows). Neck MRI showed low signal intensities (arrows) and did not show significant difference of thickness between the right and left SCM. (B) Five year old girl with left CMT showing a cord-like left SCM (arrows) and low signal intensity (arrows) on left SCM. (C) Twenty year old woman with right CMT showing atrophied cord-like right SCM (arrows) with low signal intensity (arrows) on right SCM.



## Normal SCM

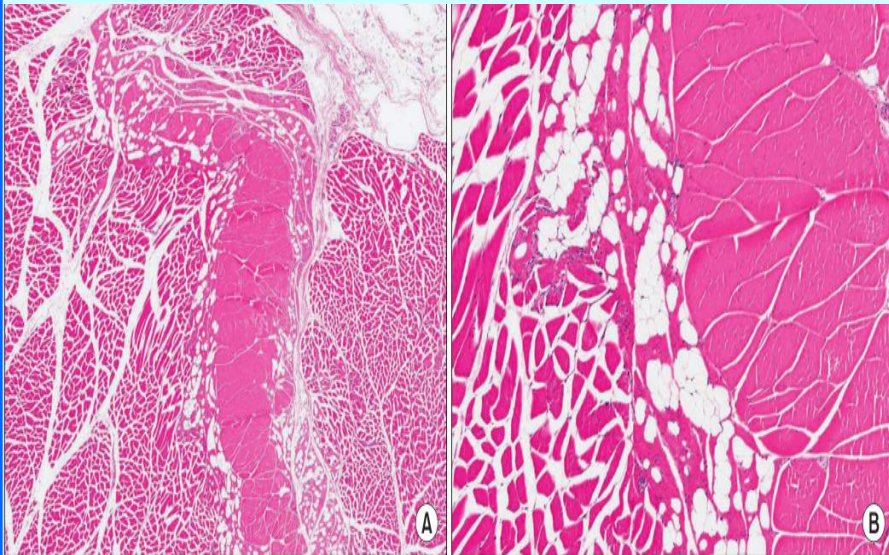


Fig. 6. The histologic findings of the normal sternocleidomastoid muscle without congenital muscular torticollis (H&E, (A)  $\times 100$ ; (B)  $\times 200$ ).

## SCM with CMT

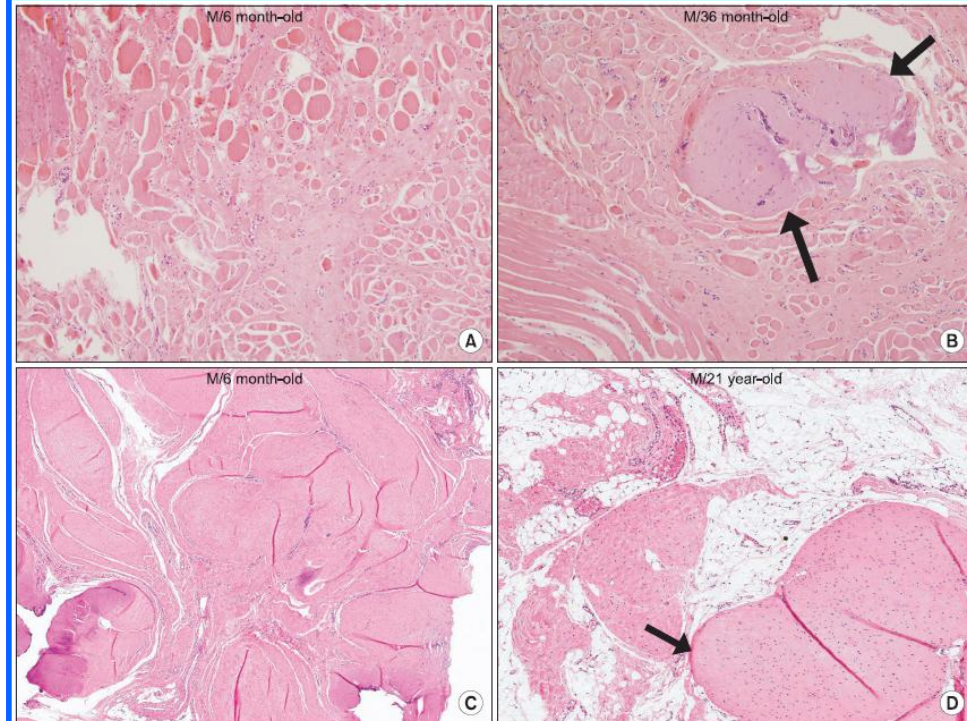


Fig. 7. The histopathological findings of the sternocleidomastoid muscle with congenital muscular torticollis. (A) Diffuse interstitial fibrosis with accompanying atrophic muscle fibers is noted (H&E,  $\times 200$ ). (B) Interstitial fibrosis with presence of aberrant tendon-like excessive dense connective 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,  $\times 40$ ).

Hwang JH, Lee HB, Kim JH, Park MC, Kwack KS, Han JD, Yim SY. Magnetic resonance imaging as a determinant for surgical release of congenital muscular torticollis: correlation with the histopathologic findings. *Ann Rehabil Med.* 2012;36:320-7.

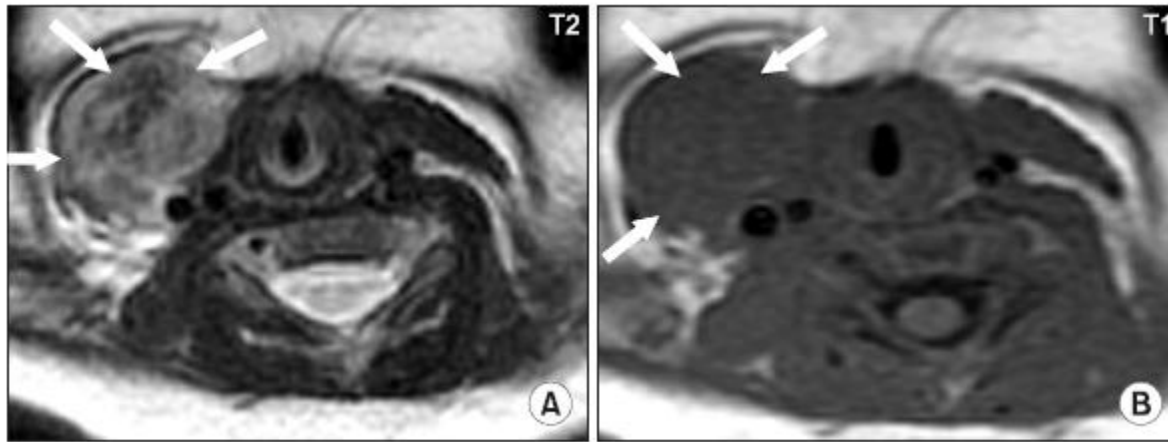
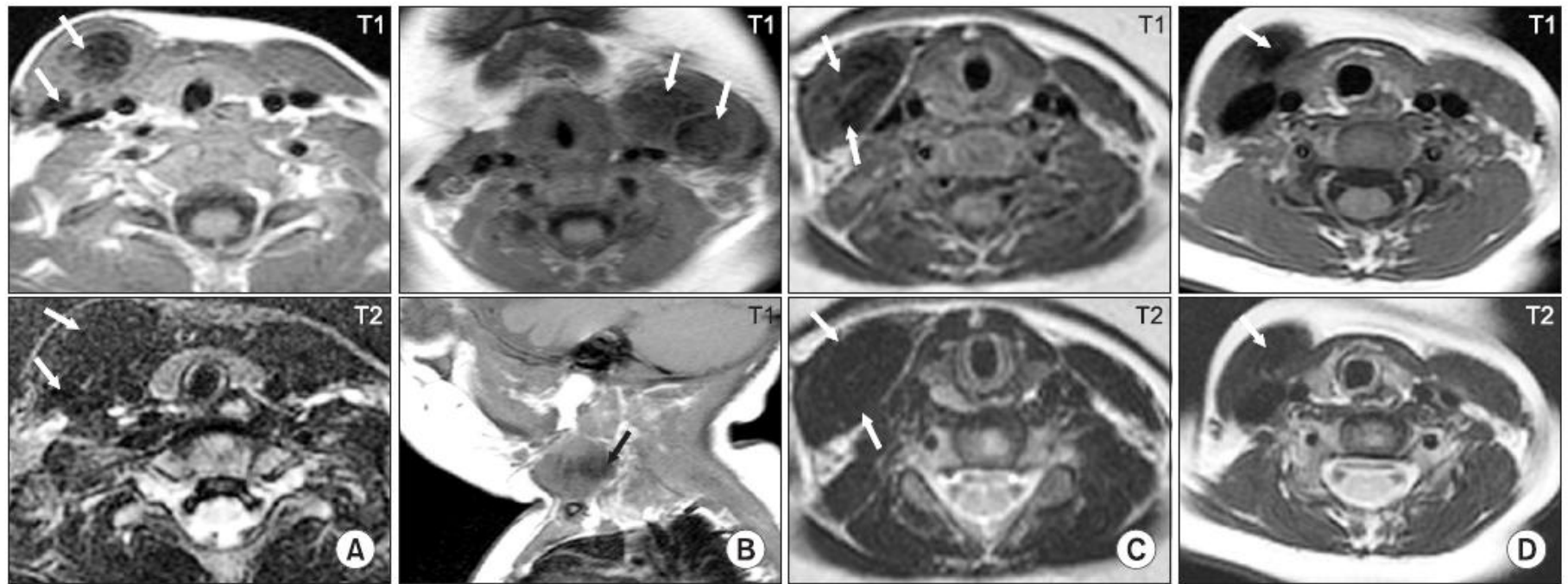


Fig. 2. The neck MRI findings of a one month old girl with right congenital muscular torticollis show high signal intensity on the (A) T2-weighted image compared to the (B) T1-weighted image (arrows).

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Hwang JH, Lee HB, Kim JH, Park MC, Kwack KS, Han JD, Yim SY. Magnetic resonance imaging as a determinant for surgical release of congenital muscular torticollis: correlation with the histopathologic findings. *Ann Rehabil Med.* 2012;36:320-7.



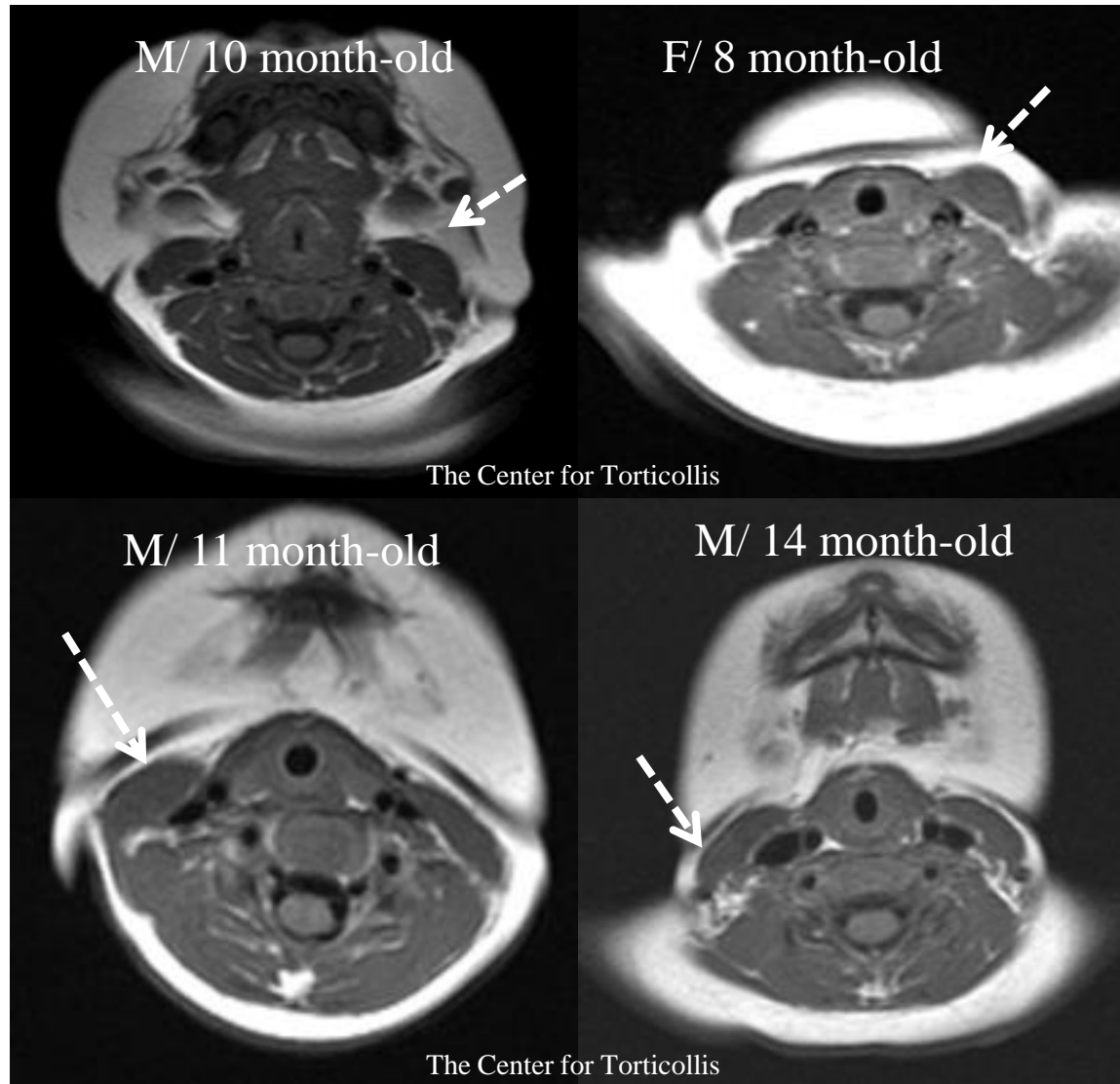


**Fig. 4.** Typical neck MRI findings of subjects who underwent surgical release for congenital muscular torticollis (CMT). (A) Eleven month old boy with right CMT showing low signal intensities on the T1- and T2-weighted axial images of both the sternal head and clavicular head (arrows) of the right sternocleidomastoid muscle (SCM). (B) Four month old girl with left CMT showing low signal intensities on the T1-weighted axial and sagittal images of the left SCM (arrows). (C) Six month old girl with right CMT showing low signal intensities on both the T1- and T2-weighted axial images of the right SCM (arrows). (D) Eighteen month old girl with right CMT showing low signal intensity on both the T1- and T2-weighted axial images of the right SCM (arrows).

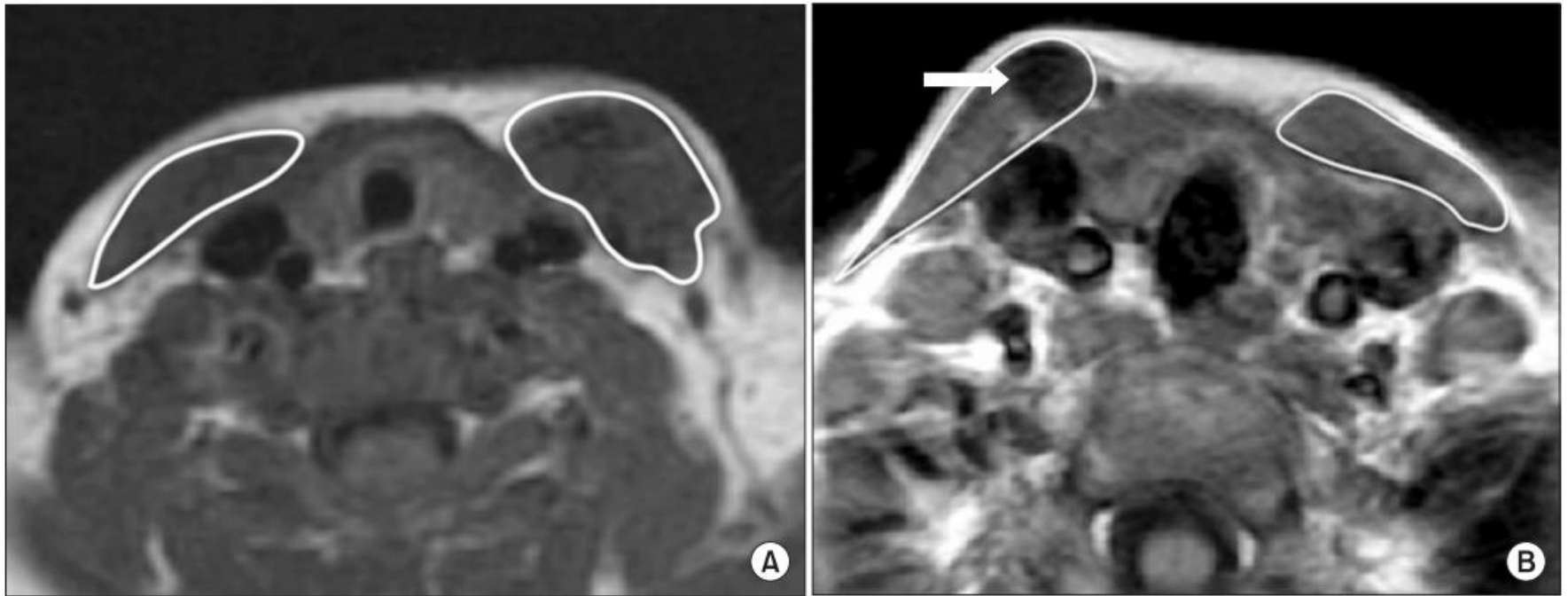
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The neck MRI findings of four subjects who showed good prognosis with the stretching exercises just revealed no low signal intensity.



Hwang JH, Lee HB, Kim JH, Park MC, Kwack KS, Han JD, Yim SY. Magnetic resonance imaging as a determinant for surgical release of congenital muscular torticollis: correlation with the histopathologic findings. *Ann Rehabil Med.* 2012;36:320-7.



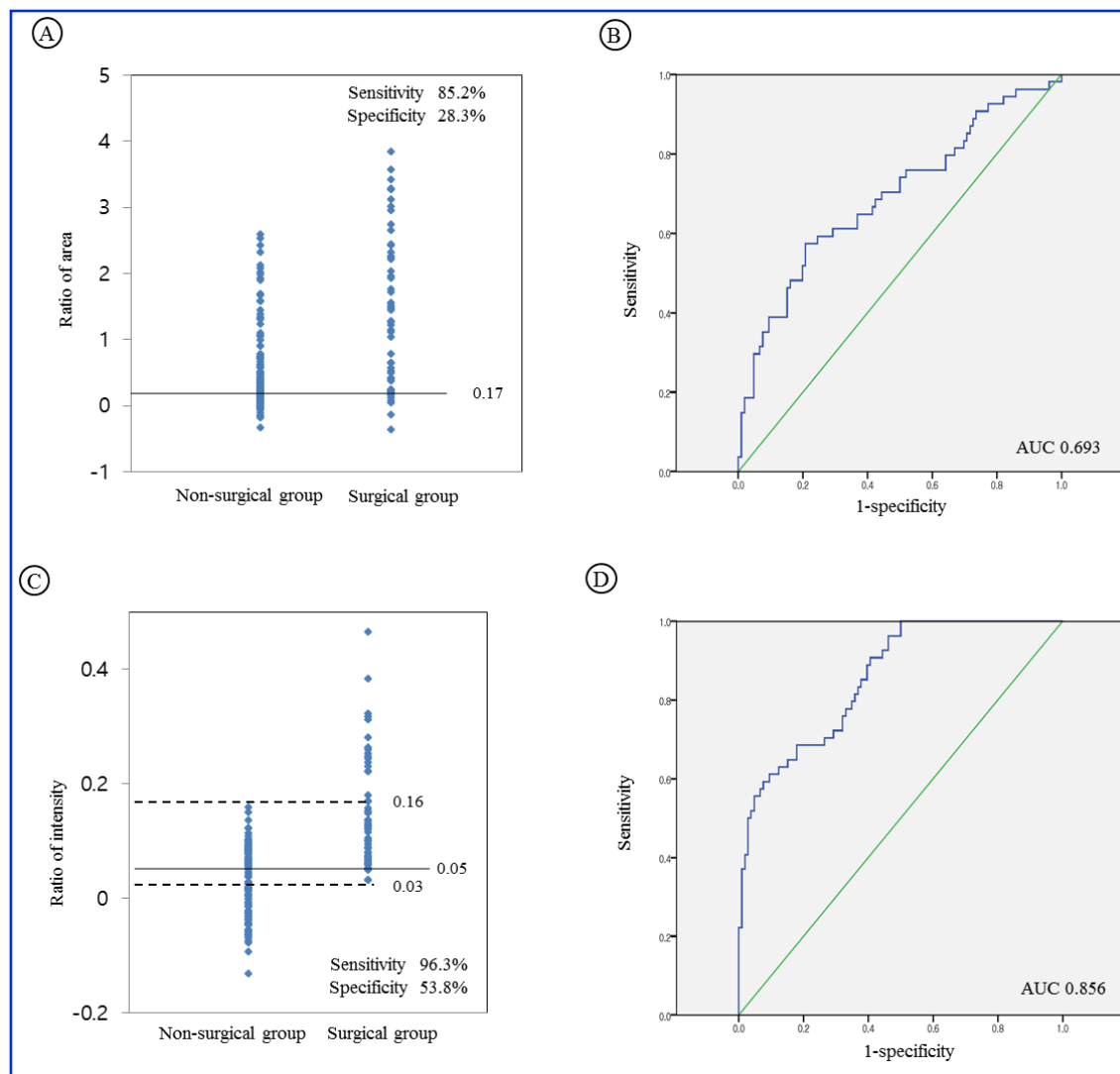
**Fig. 2.** T1-weighted magnetic resonance image of the neck of a patient with congenital muscular torticollis. (A) Methods for measuring the sternocleidomastoid muscle (SCM) area by drawing regions of interest around the SCM. (B) Methods for measuring the SCM gray color intensity (arrow) by drawing regions of interest around the SCM.

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Kim JW, Kim SH, Yim SY.

Quantitative analysis of magnetic resonance imaging of the neck and its usefulness in management of congenital muscular torticollis.

Ann Rehabil Med 2015; 39: 294-302.



Dot histogram of quantitative magnetic resonance imaging findings of the neck affected by congenital muscular torticollis (CMT) for both the non-surgical and surgical groups.

Kim JW, Kim SH, Yim SY.

Quantitative analysis of magnetic resonance imaging of the neck and its usefulness in management of congenital muscular torticollis.

Ann Rehabil Med 2015; 39: 294-302.



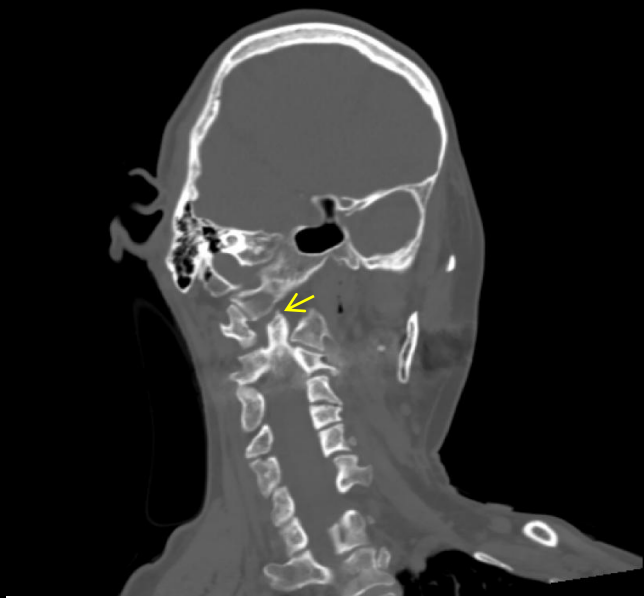
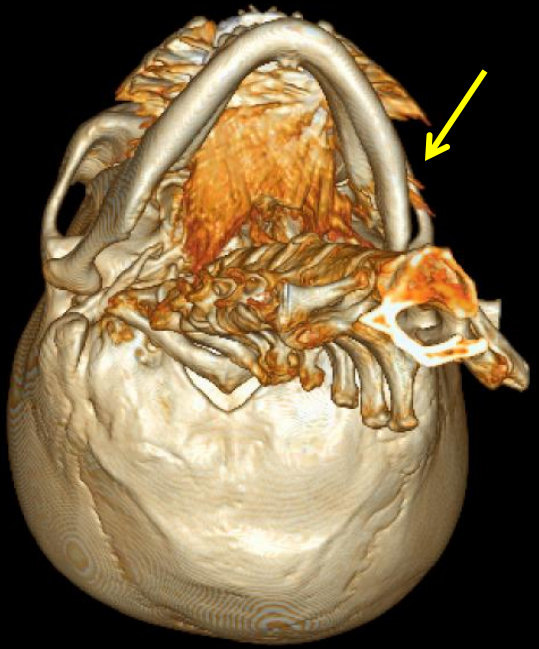
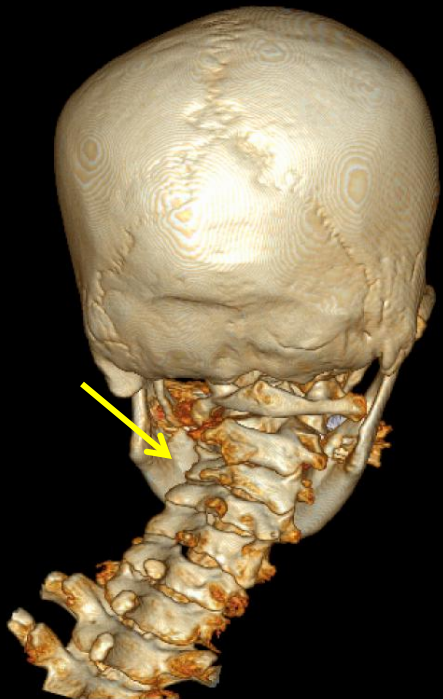
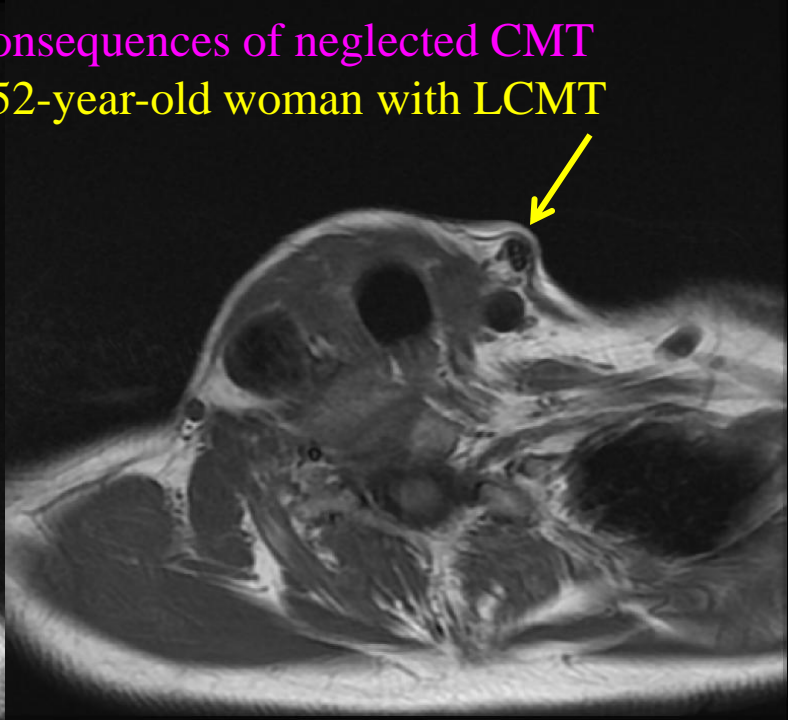
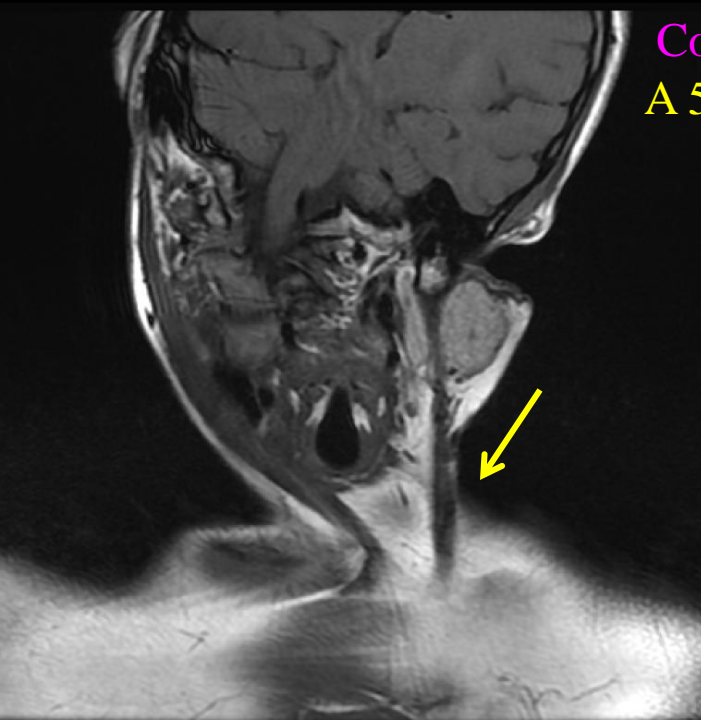
## Neglected CMT



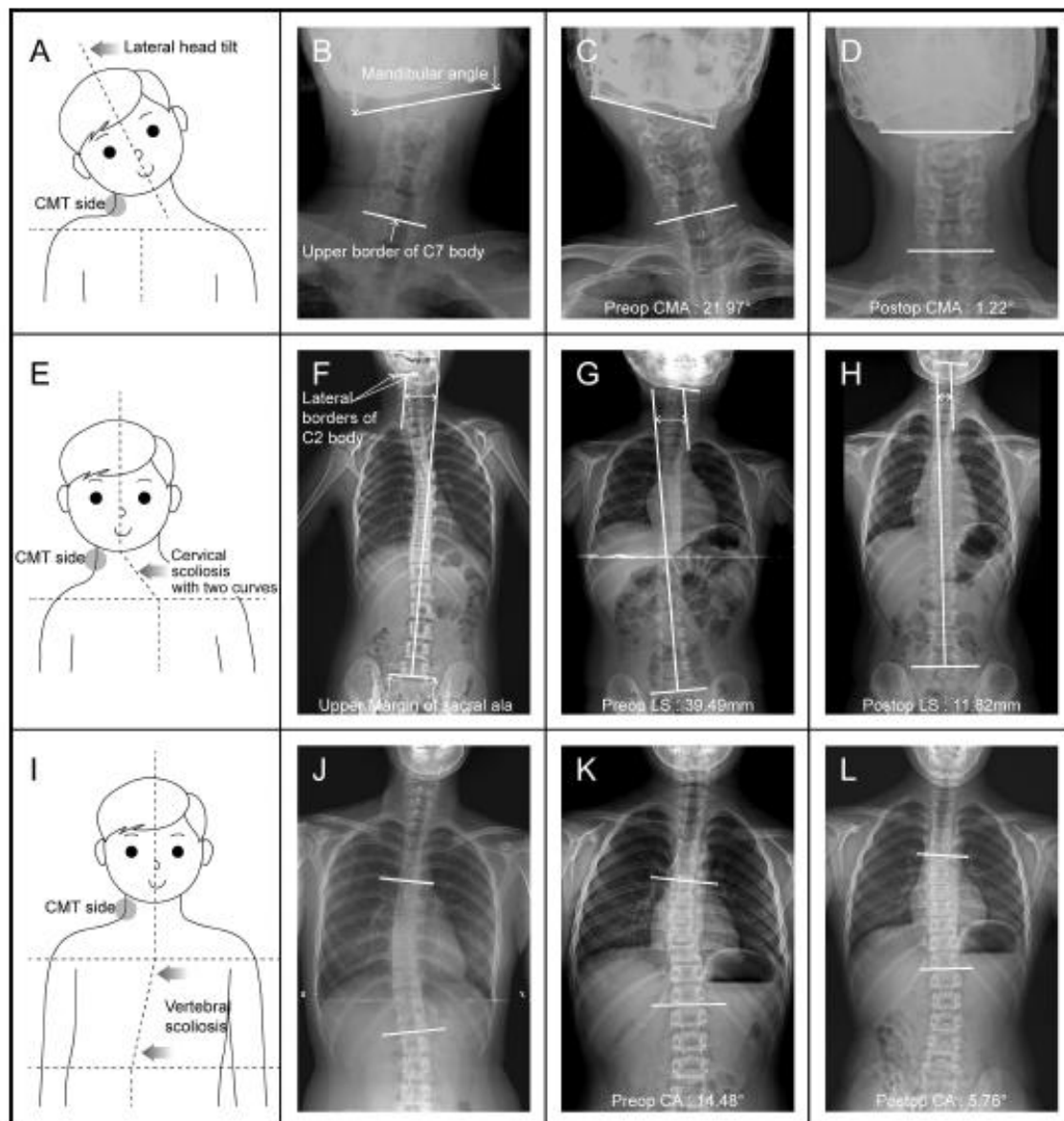
아주대학교병원 재활의학과  
사경센터 · 아동재활클리닉

Physical Medicine & Rehabilitation, Ajou University Hospital  
The Center for Torticollis · The Children's Rehabilitation Clinic

Consequences of neglected CMT  
A 52-year-old woman with LCMT







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Effectiveness of surgical release in patients with neglected congenital muscular torticollis according to age at the time of surgery

Ann Rehabil Med (in press)

**Table 2. Study Characteristics and Patient Populations of 12 Interrupted Time Series Studies Analyzed in this Review**

Study <sup>reference number</sup>	Study design	Number of patients	Age at the time of surgery (years)	Follow-up period (years)	Type of surgical intervention
Lim 2014 <sup>8</sup>	retrospective	37	27.4	2.4	Unipolar or bipolar release
Patwardhan 2011 <sup>44</sup>	retrospective	12	24.0	2.96	Bipolar release and Z plasty
Sudesh 2010 <sup>45</sup>	retrospective	14	13.4	3	Bipolar release
Omidi-Kashani 2008 <sup>46</sup>	prosepective	14	21.9	2.5	Bipolar release
Shim 2004 <sup>17</sup>	retrospective	32	14	3.25	Unipolar or bipolar release
Arslan 2002 <sup>47</sup>	retrospective	12	11.75	3.5	Bipolar release
Chen 2000 <sup>48</sup>	retrospective	18	11	5	Unipolar or bipolar releases and/or Z plasty
Akazawa 1993 <sup>49</sup>	retrospective	4	8.8	9.5	Unipolar releases
Minamitani 1990 <sup>13</sup>	retrospective	19	11.2	2.2	Unipolar releases
Itoi 1990 <sup>50</sup>	retrospective	15	10.6	7.7	Unipolar or bipolar release
Tse 1987 <sup>51</sup>	retrospective	14	11.89	3.63	Unipolar or bipolar release
Ling 1976 <sup>9</sup>	retrospective	29	≥9	No mention	Unipolar or bipolar release
Total	1 prospective 11 retrospective	220	-	-	

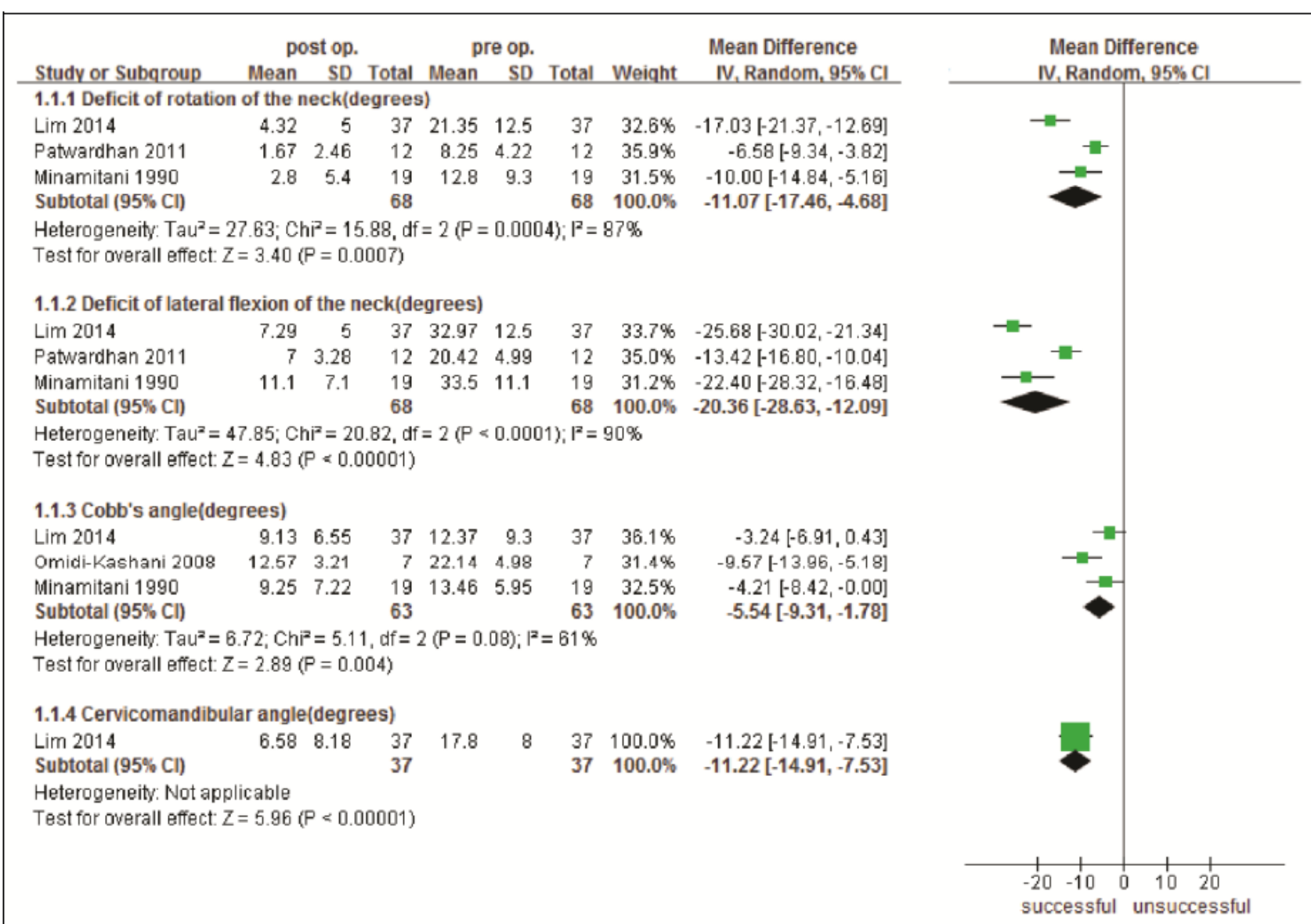
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Kim HJ, Ahn HS, Yim SY. Effectiveness of surgical treatment for neglected congenital muscular torticollis: A systematic review and meta-analysis. *Plast Reconstr Surg.* 2015;136:67e-77e.

**Table 6. Meta-Analysis of the Successful Result of Surgical Treatment for the Neglected Congenital Muscular Torticollis**

Scoring System Reference Numbers	No. of Studies	Sample Size	No. of Successful Events	Point Estimates of Successful Event Rates (Random Effects Model)		$I^2$	Heterogeneity ( $p$ )
				Point Estimate	95% CI		
Cheng and Tang's scoring system <sup>8,17,44</sup>	3	81	81	0.98	0.91–1.00	0.00	0.84
Lee and Kang's scoring system <sup>45–48</sup>	4	58	50	0.86	0.75–0.93	0.00	0.98
Canale's scoring system <sup>49–51</sup>	3	33	28	0.81	0.63–0.92	0.00	0.37
Ling's scoring system <sup>9</sup>	1	29	18	0.62	0.44–0.78	0.00	1.00
Overall	11	201	177	0.81	0.73–0.87	46.20	0.05
Test for subgroup differences							0.00

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Kim HJ, Ahn HS, Yim SY. Effectiveness of surgical treatment for neglected congenital muscular torticollis: A systematic review and meta-analysis. *Plast Reconstr Surg.* 2015;136:67e-77e.

**Table 3. Subgroup Meta-Analysis of the Successful Result of Surgical Treatment for the Neglected Congenital Muscular Torticollis between Surgery before and after 15 Years of Age**

Scoring system	Number of studies	Number of successful event	Sample size	Point estimates of successful event rates (random effects model)		Heterogeneity (Cochran's $Q$ )	I-squared ( $I^2$ )	Heterogeneity ( $p$ -value)
				Point estimate	95% CI			
				Cheng and Tang's scoring system <sup>8,17,44</sup>	3			
Surgery $\leq$ 15 year-old <sup>17</sup>	1	32	32	0.98	0.80-1.00	0.00	0.00	1.00
Surgery $>$ 15 year-old <sup>8,44</sup>	2	49	49	0.98	0.86-1.00	0.29	0.00	0.59
Lee and Kang's scoring system <sup>45-48</sup>	4	50	58	0.86	0.75-0.93	0.20	0.00	0.98
Surgery $\leq$ 15 year-old <sup>45,47,48</sup>	3	38	44	0.86	0.72-0.94	0.19	0.00	0.91
Surgery $>$ 15 year-old <sup>46</sup>	1	12	14	0.86	0.57-0.96	0.00	0.00	1.00

CI, confidence interval

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Kim HJ, Ahn HS, Yim SY. Effectiveness of surgical treatment for neglected congenital muscular torticollis: A systematic review and meta-analysis. *Plast Reconstr Surg.* 2015;136:67e-77e.



감사합니다



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